THE UNIVERSITY *of* EDINBURGH

The Potential of Acceptance and Commitment Therapy (ACT) to Improve Outcomes in Muscle Disorders:

A Longitudinal Investigation of Psychological Flexibility and Systematic Review of ACT for Long-Term Conditions

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Doctorate in Clinical Psychology

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The University of Edinburgh

DEDICATION

To my father, mother, sisters and brother who also made this possible.

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THESIS ABSTRACT

Muscle disorders are chronic, progressive conditions, the majority of which are without disease modifying treatments. Quality of life (QoL) is reduced in these conditions, and alternative methods, such as psychological intervention, may offer ways to improve QoL. Previous work has suggested that aberrant illness perceptions may be influential targets for psychological interventions; however, emerging evidence suggests that psychological flexibility might offer another treatment target.

This thesis first presents a longitudinal investigation of the role of these two variables, alongside disability level, in explaining life satisfaction and mood measured four months later. Participants were recruited from charities and online communities, with data collected via online questionnaires. Here, illness perceptions and psychological flexibility, but not disability level, were cross-sectionally associated with all dependent variables. In prospective analyses psychological flexibility accounted for greater variance in life satisfaction and anxiety; while illness perceptions explained more variance in depression. However, after controlling for variance in time one dependent variables, psychological flexibility alone was predictive of life satisfaction and anxiety at time two. Therefore, psychological flexibility represents a possible influential target for psychological intervention in muscle disorders.

Acceptance and Commitment Therapy (ACT) is a psychological intervention specifically designed to improve psychological flexibility. Subsequently, the results of the empirical study imply that ACT is worthy of trial with muscle disorders. However, there has been no comprehensive review of the use of ACT in chronic disease or long-term conditions. Therefore, Chapter 2 presents a systematic review of ACT as applied to chronic disease/long-term conditions. The aims were to collate all ACT interventions

with chronic disease/long-term conditions; evaluate their quality and comment on efficacy.

Ovid MEDLINE, EMBASE and Psych Info were searched, with a further search of citing articles undertaken using Google Scholar. Studies with mental health or chronic pain populations were excluded. Study quality was then rated, with a proportion re-rated by a second researcher. Seventeen studies were included, of which: eight were randomised controlled trials (RCTs), three used pre-post designs, and seven were case studies. A broad range of applications were observed (e.g. improving quality of life and symptom control, reducing distress) across many diseases/conditions (e.g. HIV, cancer, epilepsy). However, study quality was generally low, and many interventions were of low intensity. The small number of RCTs per application and lower study quality emphasise that ACT is not yet a well-established intervention for chronic disease/long-term conditions. However, there was promising evidence for certain applications: the parenting of children with long-term conditions, seizure-control in epilepsy, psychological flexibility and possibly self-management/lifestyle.

The studies comprising this thesis suggest that, whilst psychological flexibility appears influential in muscle disorders, high-quality research into ACT interventions for chronic disease/long-term conditions is generally lacking. Therefore one cannot confidently generalise from existing studies that ACT will improve outcomes in muscle disorders. Thus an evaluation of ACT in the context of muscle disorders is now required. This should adhere to the methodological suggestions provided in the systematic review.

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Lastly, Maria-Angelica Florez-Graham you are even more beautiful now I see you here beside me; and so we keep moving forward.

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Psychological Flexibility is More Strongly Predictive of Distress and life satisfaction in People with Muscle Disorders than Illness Perceptions or Disability Level: A Prospective Longitudinal Study

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Psychological flexibility is more strongly predictive of distress and life satisfaction in people with muscle disorders than disability level or illness perceptions: A prospective longitudinal study.

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ABSTRACT

Abstract

Background: Acceptance and Commitment Therapy (ACT), which targets psychological flexibility, and illness perceptions interventions, appear to improve quality of life in chronic disease.

Purpose: To optimize interventions for muscle disorders, after controlling for disability level, the relative influence of illness perceptions and psychological flexibility on life satisfaction and distress was investigated.

Methods: Data were collected via online questionnaires. Regression analyses examined relationships between disability level, psychological flexibility (experiential avoidance, cognitive fusion and valued living) and illness perceptions, and outcomes (cross-sectionally and 4 months later).

Results: Illness perceptions and psychological flexibility, but not disability level, were cross-sectionally associated with all outcomes. In prospective analyses psychological flexibility was most strongly associated with life satisfaction and anxiety; while illness perceptions were most strongly associated with depression. After controlling for variance in time 1 dependent variables, psychological flexibility alone was predictive of life satisfaction and anxiety at time 2.

Conclusions: Targeting psychological flexibility may improve outcomes in muscle disorders.

INTRODUCTION

Muscle disorders (MDs) are a diverse group of genetic and acquired neuromuscular conditions, the majority of which are progressive (1). They principally affect muscle tissue, which translates into an insidious decline in physical functioning and mobility. Weakness in other muscle groups may cause dysarthria, dysphagia, contractures, ptosis and opthalmopariesis. Cardiac and respiratory symptoms may also be present, and problematic pain and fatigue are frequently reported (1-4). People with MDs experience reduced quality of life when compared to population norms, and mood may also be affected (5-7).

Few disease modifying treatments are available for the majority of MDs and there are no cures. Thus, most interventions seek to manage symptomatic complications. For example, physiotherapy and orthoses are often provided to enable as broad a range of movement as possible, while medication may be prescribed for pain and fatigue (1).

Emerging evidence suggests that cognitive behavioural therapies may offer an additional way to improve quality of life and mood in MDs (8); with promising results observed in a recent trial of cognitive behavioural therapy reducing fatigue (9). This, the only published trial of a psychological intervention for MDs, utilised a three-arm randomized-controlled trial (RCT) design - comparing cognitive behavioural therapy to aerobic exercise training and treatment as usual. Here aspects of cognitive therapy targeted 'dysfunctional cognitions' regarding fatigue, sleep, activity and catastrophizing, amongst a broad range of other therapeutic methods – with sessions also aimed at improving communication between people with MDs and care-givers/relatives. Results showed significant improvements in fatigue in both the aerobic exercise training and the cognitive behavioural therapy group; however, only the cognitive behavioural therapy group showed significant improvements in social

participation and sleep compared to controls. Alongside these additional benefits the cognitive behavioural therapy treatment was much shorter in duration than the aerobic exercise training, suggesting greater cost-effectiveness. However, this trial suffered from much poorer adherence to treatment in the aerobic exercise training group, which may account for the differential outcomes. Indeed, the control group was treatment as usual (i.e. without a control for supportive therapy, empathic concern etc), and so it could also be argued that a placebo effect/impact of non-specific therapy factors were not adequately controlled.

This sole existing trial of a psychological intervention in MD sought to improve fatigue, but not QoL or mood. Given that these important outcomes also appear to be affected by living with MDs (5, 7), an outstanding central question for psychological intervention development is which cognitive and behavioral variables should be optimally targeted to improve quality of life or mood? The present study examines the relative importance of two possible targets for intervention: illness perceptions and psychological flexibility.

Illness Perceptions

Illness perceptions are cognitions formed in response to a health threat (10). They include beliefs about the time-course of the health threat, its consequences and if it can be cured or controlled amongst a range of other beliefs. Illness perceptions are theorized to influence the methods that a person will use to manage or cope with their illness; the success or failure of these methods will then affect quality of life and mood (11). In agreement with this assumption, aberrant illness perceptions are often associated with negative outcomes in chronic disease (12, 13). Subsequently, interventions which target illness perceptions have been developed. These provide

education about the disease in question and challenge inaccurate beliefs as a means to improve disease self-management (10).

Three studies have observed significant associations between illness perceptions and quality of life and mood in MDs (14-16), even after controlling for disability level. This provides promising evidence that illness perceptions are suitable targets for intervention in MDs. However, all these studies employed cross-sectional designs. This is problematic because it is conceivable that low quality of life and mood might cause aberrant illness perceptions as well as vice versa. Analyses using prospective designs would allow greater understanding of the direction of this relationship.

Psychological Flexibility

In contrast, Acceptance and Commitment Therapy (ACT) (17) proposes that one's relationship with their thoughts may have a greater influence over behavior than the content of these thoughts (for example, illness perceptions). ACT posits that a process called psychological flexibility has a key influence on quality of life and mood, especially in the face of adversity such as that presented by chronic illness. Psychological flexibility is defined as: being open, aware and in contact with present moment, flexibly pursuing behaviours which are in line with one's chosen values (18). It can be broken down into six overlapping and interdependent sub-processes: being aware of what is personally important (values) on an ongoing basis (present-moment-focus); taking steps towards these values (committed action) whilst accepting subsequent uncomfortable private experiences such as pain, embarrassment or negative thoughts (experiential acceptance); within this, being able to see thoughts as transient mental events (cognitive defusion) which are separate from the person who is doing the thinking (self-as-context).

Psychological flexibility has shown strong relationships with a range of outcomes in other conditions, such as pain interference, mood and functioning in chronic pain patients (19) and adjustment in other neuromuscular conditions (20). There is emerging evidence that psychological flexibility may also be influential in MDs. For example, one study observed that willingness to experience musculoskeletal pain (experiential acceptance) when this facilitated valued-activity, was negatively associated with depression, while engagement in valued activity (valued-living) was negatively associated with depression and positively associated with quality of life (21). Another study observed that fear of experiencing discrimination (cognitive fusion) had a greater negative correlation with quality of life than did actual experienced discrimination (22). To our knowledge the prospective influence of psychological flexibility on quality of life or mood has not yet been investigated in MDs. Psychological flexibility is postulated to influence one's ability to align their behaviours with deeply held values and stay in contact with the present moment as opposed to being drawn into unhelpful focus on worries or fears. This despite unpleasant and challenging experiences - such as ongoing pain, fatigue, illness related worries- which may occur as one lives with a MD. Thus, since psychological flexibility is theorized to impact on one's ability to undertake personally meaningful activities (both personal and illness-related), as opposed to solely one's ability to manage their illness it is a more general, and possibly more influential , process than disease self-management. Thus psychological flexibility may prove more influential on outcomes than illness perceptions.

The Present Study

This study aimed to assess the cross-sectional and prospective influence of validated measures of illness perceptions, and facets of psychological flexibility

(experiential avoidance, cognitive fusion and valued living) on life satisfaction and mood. As described in the preceding paragraph above these facets of psychological flexibility were chosen based on existing studies (21, 22), since they appear to be key processes in successful living with MDs, (8, 21).

As all available brief quality of life measures were confounded by items capturing physical functioning, a measure of life satisfaction was instead used in the present study (Satisfaction With Life Scale (23)). Life satisfaction is conceptually commensurate to quality of life, defined as: "…a global evaluation by the person of his or her life." (24), p150. These outcomes life satisfaction (i.e. QoL) and mood were deemed important since both (5, 7) are reduced in MD, and are appropriate treatment targets for forthcoming psychological intervention (see Graham et al., (2015)(8))

We hypothesized that both illness perceptions and facets of psychological flexibility would be strongly correlated with life satisfaction, anxiety and depression. We expected psychological flexibility to have a stronger prospective influence on life satisfaction and mood measured four months later.

METHODS

Participants and Procedure

Participants were recruited via a news story published on the Muscular Dystrophy Campaign (United Kingdom) website. This contained a link to a website which comprised information regarding the study, a consent form and the first questionnaire battery. Before giving consent potential participants assessed themselves against the inclusion/exclusion criteria. Prospective participants were eligible if they had a diagnosis of MD with duration of greater than six months and were aged 18 - 75 years. They were ineligible if: they had major active co-morbidities unrelated to MD

(such as stroke); experienced cognitive impairment or had myotonic dystrophy (Myotonic Dystrophy is associated with cognitive impairment (25)); were unable to read English; had major diagnosed active mental health co-morbidities, for example psychosis or major depression; or, were currently participating in treatment intervention studies.

Participants then gave consent, entered their e-mail address and progressed to the first questionnaire battery. This battery contained measures of dependent and independent variables (listed below) and recorded demographics (MD diagnosis, age, gender, and years since they first noticed symptoms). Four months later, participants were e-mailed a link to the second questionnaire battery. This again recorded the dependent variables but omitted the independent variables (to reduce participant burden), with the exception of disability level and illness perceptions (illness perceptions data are not presented here). Favorable ethical opinion was given by the Health in Social Sciences departmental ethics committee at the University of Edinburgh.

Measures

Dependent Variables

As previously described, **The Satisfaction With Living Scale (SWLS) (23)** was used instead of a measure of QoL since it records a person's estimation of how their experience of life meets their expectations, but is not confounded by items related to disease severity or symptoms. It is a seven item, uni-dimensional measure of life satisfaction. Scores can range from 0-35, with higher scores indicating greater life satisfaction. It showed good internal consistency in a previous study with people with MDs (26), and strong psychometric properties (discriminant validity, factor structure, and concurrent validity) in a range of groups, including chronic illness groups (24, 27).

In the present study the SWLS showed a very good level of internal consistency (α = .88).

The Generalised Anxiety Disorder 7-item Scale (GAD-7) (28) was chosen as a measure of anxiety not only because it is well validated (29), but also brief – just seven-items- meaning that it represents a low participant burden. Scores can range from 0-21, with higher scores indicating more severe anxiety. Psychometric evaluation in a large sample of people with another neuromuscular disease (multiple sclerosis) revealed reliability and internal validity (30). The internal consistency of this measure was excellent (α = .90).

The 9-item version of the Patient Health Questionnaire (PHQ-9) (31) is a (nine item) measure of depression. As with the GAD-7 it is well validated and brief (31), and has shown good psychometric properties in a range of other chronic illnesses e.g. (32, 33). Though to our knowledge it has not yet been applied in studies with MD populations. Scores can range from 0-27, with higher scores indicating more severe depression. The internal consistency of this measure was also very good (α = .86).

Independent Variables

The Stanford Health Assessment Questionnaire – Disability Index (HAQ-DI) (34) was chosen as it is a well-validated measure of impairment in physical functioning, or disability level (35), which is not confounded by items measuring mood or psychological constructs. It contains eight activity domains (dressing, arising, eating, walking, hygiene, reach, grip, and activities), but does not record impairment in social or emotional functioning. There are various ways to score the HAQ-DI; we used the alternative scoring method which does not take into consideration the use of aides and devices and averages the domains to give one total score (range 0 – 3)(36). Higher scores indicate greater functional impairment. Previous studies with chronic disease populations (35) (including with MD (14, 16)) have observed good reliability, and

validity. In the present study, internal consistency of the sub-scales ranged from α = .71 to α = .89.

The Brief Illness Perception Questionnaire (Brief IPQ) (37) was used to capture illness perceptions. It was chosen as it is a brief measure of illness beliefs, not confounded by aspects of psychological flexibility. In the present study eight of nine possible domains were utilized. Scores on each domain can range from 0 -10. Higher scores indicate a stronger belief that: MD has many symptoms (identity), it will be chronic as opposed to acute (timeline acute/chronic), it has many consequences (consequences), it can be controlled by ones behavior (personal control) or by treatment (treatment control), it is understandable (illness coherence) or it causes distress (emotional representation) and concern (concern). These domains can be summed to give a total score, which quantifies the extent to which the illness is viewed as threatening (37). In an initial validation study the Brief IPQ showed good test- retest reliability and concurrent validity (37); however, a more recent qualitative study using cognitive interviewing, has called into question the content validity of the measure (38). In the present study the internal consistency for the combined score of the Brief IPQ was very poor (Cronbach's α = .52). Thus exploratory factor analyses were undertaken using principal components analysis (varimax rotation) (see Appendix i). The number of factors extracted was based on Kaiser's criterion (39) and inspection of the scree plot (40). This yielded three latent variables; however, only one showed acceptable internal and conceptual consistency (Cronbach's α = .71). This variable comprised the consequences, identity, emotional representation and concern domains; thus, it appears to capture the level of threat represented by MD - henceforth called IPQ Threat.

The Cognitive Fusion Questionnaire (CFQ) (41) is a single-domain measure of cognitive fusion: "the tendency for behaviour to be overly regulated and influenced by cognition, compared to other sources of behavioral influence"(41). It has seven

items, and scores can range from 7-49, with a higher score indicating greater cognitive fusion. Here, internal consistency was excellent (α = .95). The CFQ is a new measure; however, initial validation in neuromuscular disease (multiple sclerosis) showed acceptable psychometric properties and confirmed the unidimensional factor structure (41).

The Acceptance and Action Questionnaire (AAQ)(42) is a uni-dimensional questionnaire which measures experiential avoidance, defined as: a regulatory strategy comprising attempts to control or avoid unpleasant thoughts, feelings and/or bodily sensations (43). The AAQ consists of nine items. Scores can range from 7-63, with higher scores indicating greater experiential avoidance. A large validation study with people with a range of chronic diseases has shown acceptable validity (44). In the present study the internal consistency was acceptable (α = .73). A newer version of the AAQ (45) was not used as it is designed to capture all facets of psychological flexibility within one scale, while the present version is focused on recoding experiential avoidance alone.

The Engaged Living Scale (ELS) (46) is a 16 item measure of valued living, specifically derived from the engaged response style of valued-living implicit in ACT. It has a range of 0-80, with higher scores indicating greater valued-living. It is a newer measure of valued living, it was chosen because of good face validity for the construct of valued-living. In a recent validation study it has shown excellent internal consistency and concurrent validity (46). Indeed, in the present study excellent internal consistency was apparent (Cronbach's α = .94).

Data analyses plan and statistics

Three regression analyses were undertaken for each dependent variable. The first regression analysis assessed the cross-sectional relationship between dependent and

independent variables both measured at time one. The second, the prospective association between time 1 variables and dependent variables measured 4 months later (time 2). The third, assessed the relationship between independent variables measured at time 1 and dependent variables at time 2 while controlling for variance in the dependent variable measured at time 1. Controlling for time 1 variance means that remaining variance is likely to be unique to the time 2 dependent variable. Thus, it can be inferred that variance in the T1 independent variable is antecedent to unique variance arising in the dependent variable at T2. Unlike in previous studies (14, 16), this allows one to investigate a direction of relationship, with data not confounded by cross-sectional relationships between dependent and independent variables (as would be the case if T1 variance in the dependent variable is not controlled). Change scores were not used since there were only two measurement points in the study (as opposed to three or more), meaning that variance in the independent variable would not be antecedent to change. However, there are two alternative methods which would allow one to account for T1 variance (a., absolute change with T1 score included as a covariate; b., residualized T2 score as the dependent variable) (47). In a detailed review, Dalecki & Willits (1991) suggest that all of these 'result in identical interpretations of the relationship between substantive variables and change', p126 para 1.

In all regression analyses independent variables were entered in the following steps: disability level (HAQ-DI), followed by illness perceptions (IPQ threat), then facets of psychological flexibility (valued-living [ELS]; cognitive fusion [CFQ]; experiential avoidance [AAQ]). In the regressions which controlled for time 1 levels of the dependent variable, the time 1 dependent variable was entered as a preliminary first step.

A power calculation, undertaken using the G-Power program (48) assuming a medium effect size and a power (I – β err prob) of 0.80, estimated a sample size of N = 98 were required to complete both questionnaires.

RESULTS

Description of cohort

At time 1, 191 participants completed all items. Of this group, 137 (71.72%) completed the second questionnaire battery. Only those completing both batteries were included in the analyses. Mann-Whitney U Tests (used due to unequal sample sizes and non-normality) and chi-square tests, corrected for multiple comparisons, revealed no significant differences between those who completed both questionnaires batteries and those who completed just battery one. The main included MD diagnoses groups were: limb-girdle muscular dystrophy (*N*=42); facioscapulohumeral muscular dystrophy (*N* = 28); inclusion body myositis (*N* =20); Becker muscular dystrophy (*N* =13) polymyositis and dermatomyositis (*N* = 7). The remainder (*N* =27) had a range of other MDs, including: oculopharyngeal muscular dystrophy, Emery-Dreifuss muscular dystrophy, and Bethlem myopathy. Upon entry into the study, the average age of participants was 46.74 years (*SD* = 13.56), while the average number of years with symptoms was 23.19 years (*SD* = 14.00). The sample comprised more females (*N* = 80) than males (*N*=57).

Preliminary Data Analyses

Life satisfaction significantly worsened over the period of study, (T1 Mean: 18.54 [*SD* = 7.79], T2 Mean: 17.01 [*SD* = 7.52] $t(136) = 3.23$; $p < .01$, $d = .21$) but no significant changes in anxiety ($Z = -.36$, p = .72, $d = .04$), or depression ($Z = -.17$, $p = .86$, $d = .05$) were observed (Table 1). Disability level significantly increased over this

time-period (Z = -2.38, p = .02, d = .31), though this increase was small (Median [*SE*] T1 = 2.13 [0.72], Mean [*SD*] T1 = 2.02[0.78]; Median [*SE*] T2 = 2.13 [0.60], Mean [*SD*] T2 = 2.08 [0.72]). With the exception of disability level, significant and mostly moderate to strong correlations between all variables were evident. By contrast, disability level was only significantly associated with illness perceptions (IPQ threat) (Table 1.1).

Table 1.1. Means *(standard deviations) and inter-correlations (Pearson's r) between key study variables[a]

	M (*SD*)	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.
1. SWLS T1	18.54 (7.79)		-.49**	-.52**	-.11	-52**	-.57**	.38**	.74**	.72**	-.43**	-.40**
2. GAD-7 T1	4.31 (4.4)			.79**	.02	.50**	.53**	.60**	- .56**	-.38**	.75**	.67**
3. PHQ-9 T1	6.58 (5.36)				.08	53**	.53**	.50**	-.56**	-.50**	.65**	.83**
4. HAQ-DI T1	2.02 (0.78)					.23*	.01	.06	.03	-.10	-.07	.06
5. IPQ Threat T1	26.73 (7.66)						.53**	.46**	-.44**	-.44**	.40**	.43**
6. AAQ T1	32.88 (8.90)							.58**	-.66**	-.51**	.50**	.48**
7. CFQ T1	19.96 (9.56)								-.53**	-.23**	.52**	.43**
8. ELS T1	54.50 (13.37)									.56**	.55**	-.45**
9. SWLS T2	17.01 (7.52)										-.34**	-.44**
10. GAD-7 T2	4.23 (4.28)											.68**
11. PHQ-9 T2	6.48 (5.36)											

*Significant association at less than or equal to p =.05.
**Significant association at less than or equal to p=.01.
[a]Where appropriate, transformed variables were used; presented means (SD) are for untransformed variables.

AAQ = Acceptance and Action Questionnaire (experiential avoidance); CFQ = Cognitive Fusion Questionnaire (cognitive fusion); ELS = Engaged Living Scale (valued-living); GAD-7 = Generalized Anxiety Disorder 7 item Scale (anxiety); HAQ-DI = Health Assessment Questionnaire – Disability Index (disability level); IPQ Threat = Brief Illness Perception Questionnaire Threat Scale (illness perceptions); PHQ-9 = Patient Health Questionnaire 9 item Scale (depression); SWLS= Satisfaction with Life Scale (life satisfaction)

Regression analyses

The assumptions for regression were met, with some exceptions; HAQ-DI, CFQ, GAD 7 and PHQ-9 variables were significantly skewed (Appendix ii). These variables were transformed; however, this resulted in marked improved normality for just the CFQ and HAQ-DI variables. Thus, where GAD-7 and PHQ-9 were the dependent variables, robust regression procedures -with 5000 bootstrapped samples- were also employed. Given that the achieved sample size exceeded that a priori sample size calculation based on a medium effect size, the regression analysis should be able to detect a small-to-medium effect size.

In the cross-sectional analyses illness perceptions (ΔR^2 = 0.26-0.27, p <.001) and most facets of psychological flexibility (ΔR^2 = 0.17-0.43, p <.001) explained significant proportions of variance in all dependent variables (Tables 1.2-1.4, left section; Appendix iii). However, notably experiential avoidance did not explain significant proportions of variance in any cross-sectional regression (β =-.10-.08, p = .23-69). In the prospective analyses (Tables 1.2-1.4, middle section; Appendix iii) facets of psychological flexibility accounted for more variance in life satisfaction (ΔR^2 = 0.23, p <.001) and anxiety (ΔR^2 = 0.22, p <.001) measured 4 months later, while illness perceptions explained more variance in depression (ΔR^2 = 0.19, p <.001) measured 4 months later. In the prospective analyses which controlled for time 1 variance in the dependent variable (Tables 1.2-1.4, right section; Appendix iii) most of the variance in each time 2 dependent variable was explained by the respective time 1 dependent variable (ΔR^2 = 0.52-0.68, p <.001). Nonetheless psychological flexibility explained small but significant proportions of additional variance in life satisfaction (ΔR^2 = 0.04, p = .01) and anxiety (ΔR^2 = 0.03 p = .04); these proportions of variance are roughly analogous to a small-to-medium effect size for change. Experiential avoidance (β = -.22, p = .01) was negatively associated with life satisfaction; while cognitive fusion (β =.22,

p =.01) was positively associated with this outcome. No one facet of psychological flexibility was a significant predictor of anxiety, rather a combination of the included variables predicted a significant proportion of variance. Illness perceptions were not predictive of variance in any of these regressions (ΔR^2 = 0.00-0.01, p = .17-.99) and no independent variables were predictive of variance in depression. Disability level did not explain a significant proportion of variance in any cross-sectional (ΔR^2 = - 0.01-0.01, p =.33-80) or prospective regression (ΔR^2 = 0.00-0.01, p =.14-86) for any of the dependent variables.

Table 1.2. Three regressions showing the cross-sectional and prospective influence of independent variables on life satisfaction (SWLS)

A.Crossectional: all variables at T1				B. Prospective: Independent variables at T1; Dependent variables at T2				C. Prospective: Independent variables at T1; Dependent variables at T2 controlling for T1.				
	Step 1	**Step 2**	**Step 3**		**Step 1**	**Step 2**	**Step 3**		**Step 1**	**Step 2**	**Step 3**	**Step 4**
								SWLS T1	.72**	.72**	.67**	.59*
HAQ-DI	-.11	.01	-.08	**HAQ-DI**	-.10	.01	-.07	**HAQ-DI**		-.02	.00	-.02
IPQ-Threat		-.52**	-.22**	**IPQ-Threat**		-.45**	-.23**	**IPQ-Threat**			-.10	-.10
AAQ			-.10	**AAQ**			-.28**	**AAQ**				-.22**
CFQ			.13	**CFQ**			.30**	**CFQ**				.22**
ELS			.66**	**ELS**			.45**	**ELS**				.06
ΔF	1.68	47.92	39.22	***ΔF***	1.25	32.34	17.65	***ΔF***	147.31	0.07	1.95	3.65
ΔR²	.01	.26**	.34**	***ΔR²***	.01	.19**	.23**	***ΔR²***	.52**	.00	.01	.04*
95% CI *ΔR2*	-.02 - .04	.13 - .38	.22 - .46	***95% CI ΔR2***	-.02 - .04	.07 - .30	.11 - .34	**95% CI *ΔR2***	.40 - .63	-	-.02 – 0.04	-.02 - .10

*Significant association at less than p =.05.
** Significant association at less than or equal to p=.01.

AAQ = Acceptance and Action Questionnaire (experiential avoidance); CFQ = Cognitive Fusion Questionnaire (cognitive fusion); ELS = Engaged Living Scale (valued-living); GAD-7 = Generalized Anxiety Disorder 7 item Scale (anxiety); HAQ-DI = Health Assessment Questionnaire – Disability Index (disability level); IPQ Threat = Brief Illness Perception Questionnaire Threat Scale (illness perceptions); PHQ-9 = Patient Health Questionnaire 9 item Scale (depression); SWLS= Satisfaction with Life Scale (life satisfaction)

Table 1.3. Three regressions showing the cross-sectional and prospective influence of independent variables on anxiety (GAD-7)[b]

A. Crossectional: all variables at T1				B. Prospective: Independent variables at T1; Dependent variables at T2				C. Prospective: Independent variables at T1; Dependent variables at T2 controlling for T1.				
	Step 1	**Step 2**	**Step 3**		**Step 1**	**Step 2**	**Step 3**		**Step 1**	**Step 2**	**Step 3**	**Step 4**
								GAD7 T1	.75**	.75**	.71**	.59*
HAQ-DI	.02	-.10	-.04	**HAQ-DI**	-.07	-.17*	-.11	**HAQ-DI**		-.09	-.10	-.08
IPQ-Threat		.52**	.20*	**IPQ-Threat**		.44**	.12	**IPQ-Threat**			.09	.00
AAQ			.03	**AAQ**			.06	**AAQ**				.05
CFQ			.38**	**CFQ**			.31**	**CFQ**				.08
ELS			-.23*	**ELS**			-.27**	**ELS**				-.14
ΔF	.06	46.18	19.75	***ΔF***	0.64	30.09	16.53	***ΔF***	168.07	2.20	1.00	2.78
ΔR²	.00	.26**	.23**	***ΔR²***	.01	.18**	.22**	***ΔR²***	.56**	.01	.00	.03*
95% CI ΔR2	-	.13 - .38	.11 - .35	***95% CI ΔR2***	-.02 - .04	.07 - .29	.10 - .34	***95% CI ΔR2***	.45 - .67	-.02 - .04	-	-.02 - .08

*Significant association at less than p =.05.
** Significant association at less than or equal to p=.01.
[b] Bootstrapped p-values reported.

AAQ = Acceptance and Action Questionnaire (experiential avoidance); CFQ = Cognitive Fusion Questionnaire (cognitive fusion); ELS = Engaged Living Scale (valued-living); GAD-7 = Generalized Anxiety Disorder 7 item Scale (anxiety); HAQ-DI = Health Assessment Questionnaire – Disability Index (disability level); IPQ Threat = Brief Illness Perception Questionnaire Threat Scale (illness perceptions); PHQ-9 = Patient Health Questionnaire 9 item Scale (depression); SWLS= Satisfaction with Life Scale (life satisfaction)

Table 1.4. Three regressions showing the cross-sectional and prospective influence of independent variables on depression (PHQ-9)[c]

A. Crossectional: all variables at T1				B. Prospective: Independent variables at T1; Dependent variables at T2				C. Prospective: Independent variables at T1; Dependent variables at T2 controlling for T1.				
	Step 1	**Step 2**	**Step 3**		**Step 1**	**Step 2**	**Step 3**		**Step 1**	**Step 2**	**Step 3**	**Step 4**
								PHQ9 T1	.83**	.83**	.83**	.82**
HAQ-DI	.08	-.04	.02	**HAQ-DI**	.06	-.04	.01	**HAQ-DI**		-.01	-.01	-.01
IPQ-Threat		.54**	.26*	**IPQ-Threat**		.44**	.19	**IPQ-Threat**			.00	-.02
AAQ			.08	**AAQ**			.18	**AAQ**				.11
CFQ			.20*	**CFQ**			.16	**CFQ**				-.01
ELS			-.28**	**ELS**			-.16	**ELS**				.07
ΔF	0.95	50.53	13.06	***ΔF***	0.50	30.79	7.79	***ΔF***	291.06	.03	.00	.84
ΔR²	.01	.27**	.17**	***ΔR²***	.00	.19**	.12**	***ΔR²***	.68**	.00	.00	.01
95% CI ΔR2	-.02 - .04	.15 - .39	.06 - .28	***95% CI ΔR2***	-	.07 - .31	.02 - .22	***95% CI ΔR2***	.59 - .77	-	-	-.02 - .04

*Significant association at less than p =.05.
** Significant association at less than or equal to p=.01.
[c] Bootstrapped p-values reported.

AAQ = Acceptance and Action Questionnaire (experiential avoidance); CFQ = Cognitive Fusion Questionnaire (cognitive fusion); ELS = Engaged Living Scale (valued-living); GAD-7 = Generalized Anxiety Disorder 7 item Scale (anxiety); HAQ-DI = Health Assessment Questionnaire – Disability Index (disability level); IPQ Threat = Brief Illness Perception Questionnaire Threat Scale (illness perceptions); PHQ-9 = Patient Health Questionnaire 9 item Scale (depression); SWLS= Satisfaction with Life Scale (life satisfaction)

DISCUSSION

As expected the cross-sectional analyses observed both illness perceptions and psychological flexibility to be strongly related to life satisfaction, anxiety and depression; while no association between disability level and these dependent variables was noted. Prospective analyses showed that psychological flexibility at time 1 accounted for more variance in the life satisfaction and anxiety measured 4 months later, while illness perceptions explained more variance in depression measured 4 months later. The most persuasive analyses were those that controlled for variance in the time 1 variables; as the remaining variance arises at time 2 the independent variables are thus likely to be antecedent to this. Not unexpectedly, given the short latency between measurements, the time one dependent variables accounted for large proportions of the variance in the time 2 dependent variables. Nonetheless, baseline psychological flexibility, but not illness perceptions, predicted small but significant proportions of additional variance in both life satisfaction and anxiety measured four months later. However, no independent variables predicted significant proportions of variance in depression.

Thus, as the average disability level of participants increased between the two time-points, the present results suggest that psychological flexibility may buffer declines in life satisfaction or worsening anxiety as disability level increases. In contrast, illness perceptions, whilst cross-sectionally associated, showed no significant prospective influence on life-satisfaction and mood. The success of psychological flexibility in predicting later outcomes may lie in its ability to capture the cognitive-behavioural processes which might interfere with or accentuate one's ability to pursue personally meaningful activity. This focus on over-arching values as opposed to disease self-management is a key difference between the models from which illness perceptions and psychological flexibility are derived.

The lack of influence of illness perceptions may also be explained by the limited role for disease self-management in MDs, given that few available treatments exist. Illness perceptions are perhaps likely to have greater influence in diseases where ineffective self-management might strongly interfere with valued-living outside of illness (e.g. diabetes, renal disease, cardiovascular disease) (49-51).

Nonetheless these findings lend support to the argument that cognitive behavioural interventions for MDs may benefit from using an ACT framework (8). Indeed, there is a growing evidence base for ACT interventions with chronic health conditions (52-54).

Interesting relationships between aspects of psychological flexibility and later outcomes were also observed in the present study. In agreement with a study of chronic pain in MDs (Kratz et al., 2013), later life satisfaction was negatively related to experiential avoidance. However, surprisingly life satisfaction was also positively related to cognitive fusion. Cumulatively this suggests that life satisfaction benefits from a) viewing thoughts as real literal events (cognitive fusion) while also, b) showing openness to experiencing unpleasant private events, such as difficult thoughts and feelings (experiential acceptance) when doing do serves ones values. This may align with an attitude of 'stoic' acceptance – in other words, of 'just getting on with it' - which showed positive associations with quality of life in an earlier study with MDs (55).

In the present study, psychological flexibility predicted significant proportions of variance in later anxiety. This adds to the literature which suggests that psychological flexibility influences anxiety across populations (56-58). The lack of any predictor variables to explain variance in later depression, when initial depression was controlled may be explained by lack of change in depression scores over time. While it remains possible that neither psychological flexibility or illness perceptions are predictive of

depression, positive associations between psychological flexibility (56-58) and aberrant illness perceptions (10) and depression have been noted in other populations. Thus, future studies might benefit from measuring depression over a longer period of time to allow for greater variation.

Limitations

While these analyses offer the strongest evidence to date that psychological flexibility influences life satisfaction/quality of life or anxiety in MDs, we cannot rule out the influence of unmeasured variables which affect both psychological flexibility and outcome variables. In addition, due to the entry of baseline dependent variables as controls the proportions of explained variance in prospective analyses were low. Thus experimental studies which manipulate independent variables – perhaps in an intervention study - would be required to make claims regarding causality and indicate whether these small proportions of variance are clinically meaningful.

The current sample was also drawn from online communities as opposed to hospital clinics, and was a self-selecting group and may not be representative of the true population. Several exclusion criteria (e.g. cognitive impairment, psychiatric diagnoses) were used to minimize the risk of someone without capacity consenting to take part; though meeting either criteria is far from sufficient to infer that one to lacks capacity, we assumed that lack of capacity may be more likely in these groups. This conservative approach may have limited the representativeness of the sample, compared to those seen in clinic: arguably a subsequent intervention would be more likely to be applied to those with high levels of mood disturbance. Indeed, further, one could also argue that it might have reduced the amount of variation in certain outcome measures – especially mood. This may have reduced the amounts of free variance which could be

open to explanation by the independent variables. Also, independent variables themselves may have also been affected by this loss in variance – one might expect psychological flexibility to differ in groups with psychiatric diagnoses. Conversely, one further draw-back of online recruitment was that participants assessed themselves against the inclusion/exclusion criteria, and it is thus we cannot exclude the possibility that those who did not meet these criteria- indeed even those without MDs at all - may have taken part. Nonetheless, interestingly, the demographic composition of the present sample was strikingly similar (age, years with muscle disease, and range and proportion of different MDs) to an earlier sample which was drawn from UK National Health Service clinics (14).

Also, although we compared those who completed questionnaires at both time-points to those who completed them at just one time-point on all included measures and observed no significant difference between these groups, it remains possible that these groups did differ on an unmeasured variable (e.g. attachment, pain, motivation), or that by T2 their condition, mood or life satisfaction may have deteriorated greatly such that they withdrew from the study. Thus, our assumption that responses are missing at random may be inaccurate. This possibility again calls into question the generalizability of our findings. This point is particularly important given that only small (to medium) effect sizes were observed throughout. Indeed, the 95% CI around each change in R^2 was quite broad, suggesting imprecision in the resultant R2 change scores. Thus replication would be required to achieve greater certainty about these results.

Another limitation was that, due to problems with internal consistency, half of the domains from the Brief IPQ were not used. This may have occurred because several of the items (e.g. treatment control or personal control) may not be applicable to MDs, which are generally without treatment. Another explanation for this is that the negative schema proposed by Broadbent et al., (2006)(37), are not necessarily negative in MDs,

for example greater illness coherence and less permanent timeline beliefs are not necessarily indicative of positive schemata in MDs (59). It could thus be argued that potentially influential data were not included. However, the four domains which were included were those most strongly and consistently related to domains of QoL and mood in an earlier study (14), with the resultant reduced-item scale conceptually commensurate to the full-item scale (measuring illness threat).

CONCLUSION

Psychological flexibility was a stronger prospective predictor of life satisfaction and anxiety than illness perceptions. This aligns with the view that ACT may be efficacious for improving life satisfaction and anxiety in MDs.

NEXT STEPS IN THE THESIS

The results of this chapter suggest that ACT might improve outcomes in muscle disorders. A next step would be to investigate the evidence base for the use of ACT in chronic diseases and long-term conditions. This would allow us to establish how ACT has been applied in such contexts, and whether it appears to have efficacy for improving salient outcomes.

One could argue that if ACT has convincing efficacy (and safety) for QoL/life satisfaction and distress in other long-term conditions, then, since the contexts are directly comparable, it can be applied clinically in muscle disorder care. However, if there is a lack of evidence then feasibility studies and randomised controlled trials are required before it should be recommended as part of muscle disorder care.

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CHAPTER TWO

The Use of Acceptance and Commitment Therapy (ACT) in Chronic Diseases and Long-Term Conditions: A Systematic Review

TITLE PAGE*

The Use of Acceptance and Commitment Therapy (ACT) in Chronic Disease and Long-Term Conditions: A Systematic Review of Intervention Studies

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ABSTRACT

Many have suggested that Acceptance and Commitment Therapy (ACT) may be particularly effective for improving outcomes in chronic disease/long-term conditions, and ACT techniques are now being used clinically. However, review of ACT in this context is lacking, and the state of evidence has not been clearly described. This systematic review aimed to: collate all ACT interventions with chronic disease/long-term conditions; evaluate their quality; and, comment on efficacy. Ovid MEDLINE, EMBASE and Psych Info were searched. Studies with mental health or chronic pain populations were excluded. Study quality was then rated, with a proportion re-rated by a second researcher. Seventeen studies were included: eight were randomised controlled trials (RCTs), three used pre-post designs, and six were case studies. A broad range of applications were observed (e.g. improving quality of life and symptom control, reducing distress) across many diseases/conditions (e.g. HIV, cancer, epilepsy). However, study quality was generally low, and many interventions were of low intensity. The small number of RCTs per application and lower study quality emphasise that ACT is not yet a well-established intervention for chronic disease/long-term conditions. However, there was promising evidence for certain applications: parenting of children with long-term conditions, seizure-control in epilepsy, psychological flexibility and possibly self-management.

Key words: Acceptance and Commitment Therapy; systematic review; chronic disease; cancer; HIV; long-term conditions.

INTRODUCTION

Successful living with a chronic disease/long-term condition such as diabetes, HIV, cancer or brain injury (henceforth called long-term conditions) is likely to involve a range of adaptive self-management behaviours, for example: adhering to medications, or amending one's activities and diet. A parallel process of psychological adjustment may also occur, involving evaluation of the functional impact of the condition and the regulation of any resultant distress (Leventhal, Nerenz, & Steele, 1984). The recognition that a person's own self-management behaviours and emotional responses may impact on their quality of life and other meaningful outcomes, has led to the application of cognitive behavioural interventions which target beliefs, behaviours and emotional regulation to improve outcomes (Barlow, Wright, Sheasby, Turner, & Hainsworth, 2002; Graham, Simmons, Stuart, & Rose, 2014; Petrie & Weinman, 2012).

Cognitive and behavioural interventions for long-term conditions

Interventions derived from traditional cognitive therapy have been widely applied to improve distress and self-management in long-term conditions (Greer et al., 1992; Hind et al., 2014; Ismail, Winkley, & Rabe-Hesketh, 2004; Moss-Morris et al., 2013; Petrie, Perry, Broadbent, & Weinman, 2012; Safren et al., 2014). These see one's beliefs as the central process in therapy. Subsequently, they employ techniques such as verbal modification or behavioral experiments to enable participants to change aberrant beliefs about illness, the self, the future or even medication as a means to reduce distress or instigate better self-management (Halford & Brown, 2009; Petrie, Cameron, Ellis, Buick, & Weinman, 2002; Petrie et al., 2012; Petrie & Weinman, 2012).

Acceptance and Commitment Therapy is a newer form of cognitive behavioural therapy (Hayes, Luoma, Bond, Masuda, & Lillis, 2006). It differs from traditional

cognitive therapy in two key ways. First, instead of changing the content of beliefs it aims to foster psychological flexibility, which is defined as: being open, aware and in contact with the present moment; flexibly engaging in behaviours which facilitate overarching life goals (Bond, Hayes, & Barnes-Holmes, 2006). Second, it sees distress as a natural consequence of experiencing adversity; and does not explicitly aim to reduce distress, but rather to increase one's ability to undertake valued-activity in its presence. ACT uses a range of methods to engender psychological flexibility, for example: mindfulness exercises to enable one to be present moment focused; defusion exercises to change one's relationship with thoughts; and, values elicitation exercises to orientate participants to valued behaviours (McCracken, 2011).

Why might ACT have utility in long-term conditions?

Many have expressed the opinion that ACT has utility over existing psychotherapeutic models in the context of long-term conditions (Angiola & Bowen, 2013; Graham, Simmons, et al., 2014; Hadlandsmyth, White, Nesin, & Greco, 2013; Kangas & McDonald, 2011; Karekla & Constantinou, 2010; Low et al., 2012; Moitra, Herbert, & Forman, 2011; Whittingham, 2014). For example, negative illness beliefs and distress may be realistic in certain conditions at certain times. Thus, ACT's focus on instigating valued behaviours while accepting such thoughts and feelings may prove more effective than attempts to directly alter them (as per traditional cognitive therapy) (Graham, Simmons, et al., 2014; Low et al., 2012). Others have suggested that non-adherence to HIV (Moitra et al., 2011) or diabetes (Hadlandsmyth et al., 2013) medication are related to avoidance of disease-related thoughts and feelings, such as fear or shame. Therefore, ACT's focus on encouraging (experiential) acceptance in the service of meaningful behaviour may be particularly efficacious for disease self-management. Subsequently, there is emerging evidence that ACT techniques are being

adopted by clinical health professionals working with long-term conditions (Thewes et al., 2014).

The present review

The empirical status of ACT for chronic pain (Veehof, Oskam, Schreurs, & Bohlmeijer, 2011) and general mental health populations (A-Tjak et al., 2015; Öst, 2008; Swain, Hancock, Hainsworth, & Bowman, 2013; Zum & Emmelkamp, 2009) has been previously reported. While some reviews have included a very small number of studies with long-term conditions (A-Tjak et al., 2015; Gundy, Woidneck, Pratt, Christian, & Twohig, 2011; Öst, 2008, 2014), comprehensive review of ACT as applied to long-term conditions is lacking. Given the evidence of ACT's existing clinical usage in this context (Thewes et al., 2014), calls for further application (Angiola & Bowen, 2013; Graham, Simmons, et al., 2014; Hadlandsmyth et al., 2013; Kangas & McDonald, 2011; Karekla & Constantinou, 2010; Low et al., 2012; Whittingham, 2014), and the rapid rate of growth in ACT intervention studies (Öst, 2014), we present a timely review of ACT for long-term conditions. The aim was to collate all ACT applications, to accurately describe the field. Case studies were included since they give clinically-useful descriptive accounts and allow further insight into the range of applications. It has been suggested that the general quality of ACT intervention studies is low (Hofmann & Asmundson, 2008; Öst, 2008; Ost, 2014), and that ACT has limited additional value over traditional methods (Hofmann & Asmundson, 2008). Therefore, we sought to evaluate the quality of studies which use trial methodology; comment on the emerging efficacy of ACT applications; and, suggest ways to improve the quality of future intervention studies.

METHOD

Procedures

The procedures of this systematic review were informed by accepted guidelines for systematic reviewing (Khan, Ter Riet, Glanville, Sowden, & Kleijnen, 2001; Moher, Liberati, Tetzlaff, Altman, & The, 2009). Ovid MEDLINE, EMBASE and Psych Info were systematically searched from their earliest available listing to 22nd February 2015. Due to the large number of possible long-term conditions, a broad search strategy was applied. This used the key terms ACCEPTANCE AND COMMITMENT THERAPY and CONTEXTUTAL COGNITIVE BEHAVIO$. Abstracts were examined if the title suggested an intervention study with long-term conditions. To identify further relevant studies: 1) the reference sections of the included studies were examined; 2) Google Scholar was then used to search amongst articles which had cited the included studies.

Studies were included if they described an ACT intervention applied to a long-term condition. They were excluded if they: 1) were not published in English; 2) described a hypothetical intervention; 3) did not clearly use ACT techniques; 4) were undertaken with a chronic pain (since this is well reviewed elsewhere [(McCracken & Eccleston, 2003; Veehof et al., 2011)]), or mental health population (including insomnia and conversion disorders, 'functional' illness etc); 5) were designed to prevent illness in a group without a long-term condition (see Figure 1).

Study quality was then assessed using the Psychotherapy Outcome Study Methodology Rating Form (POMRF)(Öst, 2008). This 22-item measure comprises various indicators of methodological quality, for example: length of follow-up assessment, composition of comparison interventions, reliability and specificity of measures, study design, therapist training and supervision. Items are rated as 'Poor' (0

points), ‘Fair’ (1 point) or ‘Good’ (2 points), giving a maximum score of 44 points. Two items which appeared to be related to certainty of psychiatric diagnoses were removed. Therefore, in the present study, the maximum possible score was 40.

The quality of all studies was assessed by the lead author (CDG). To improve the accuracy/validity of this assessment, a sub-section (5 papers) were randomly selected (random number generator) and also rated by another researcher with qualification to PhD level (CK). A moderate level of inter-rater agreement between reviewers was observed ($k = 0.60$, $p <. 001$) (Altman, 1991). Discrepancies were discussed and reconciled; then all articles were rated again by the lead author.

Data extraction plan

Data regarding the sample characteristics, composition of the intervention and control intervention, outcome measures and indicators of efficacy (proportion of statistically significant outcomes and effect size [Cohen’s *d*]) were extracted. Where possible the effect sizes which were reported within an included publication were used. If these were not available then effect sizes were calculated via comparison of the post-intervention scores of the experimental and control group (between-groups), or comparison between pre- and post-intervention scores (within-groups).

RESULTS

The initial database search returned 1436 studies, from which 23 publications were retrieved in full. The removal of study protocols and interventions with insomnia populations left 15 remaining studies. Four additional studies were obtained from Google Scholar. However, upon closer inspection, a study which showed low fidelity to ACT, and another with a healthy population were removed. Thus 17 studies were included in the systematic review (Figure 1).

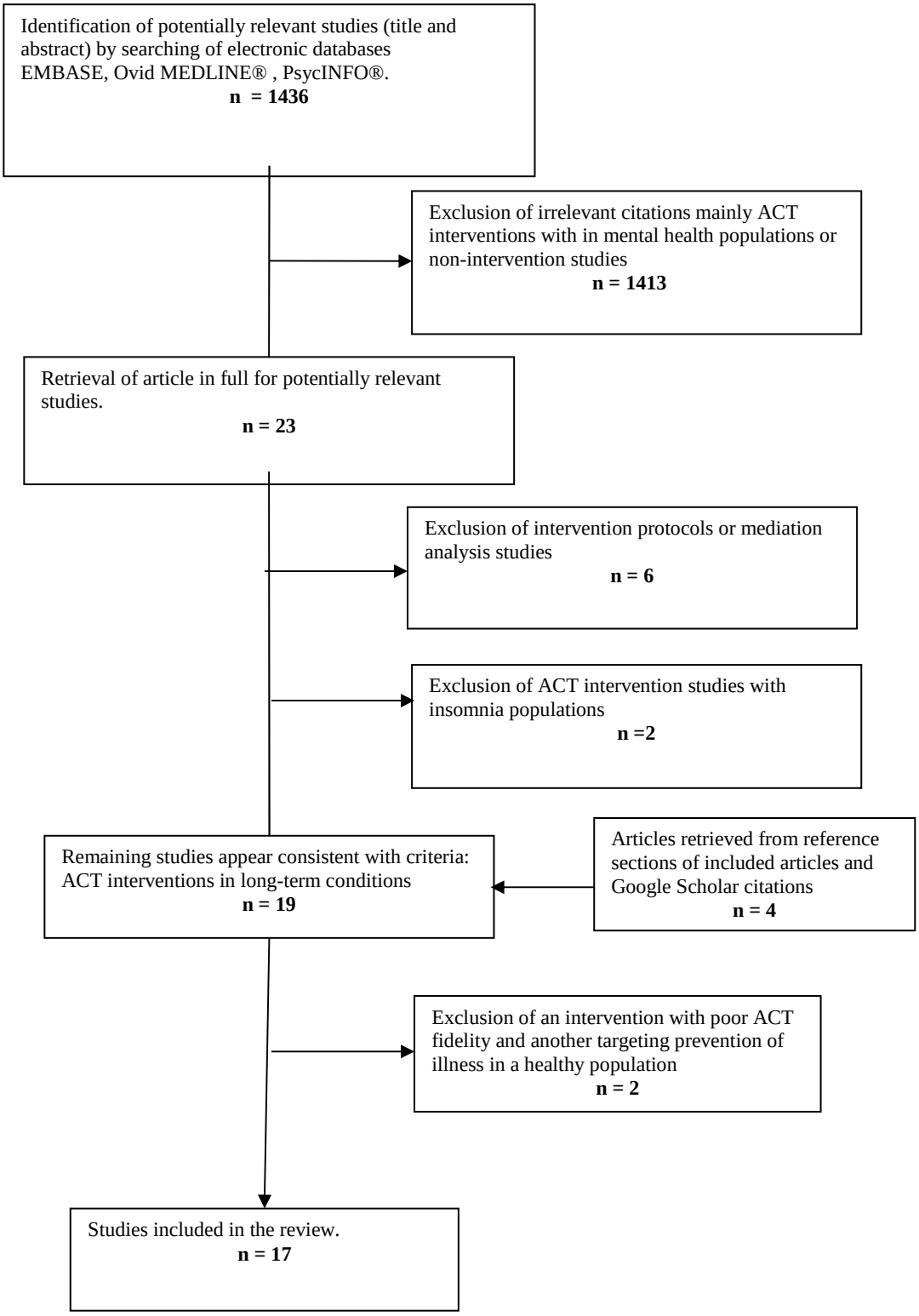

Figure 1. Flowchart showing the process of selecting studies included in the review.

Description of the included studies

Of these 17 studies: six were case studies (or case series) (Gillanders & Gillanders, 2014; Graham, Gillanders, Stuart, & Gouick, 2014; Masuda, Cohen, Wicksell, Kemani, & Johnson, 2011; Moitra et al., 2011; Nes et al., 2012; Skinta, Lezama, Wells, & Dilley, 2014); three used pre-post designs with no control group (Burke et al., 2014; Feros, Lane, Ciarrochi, & Blackledge, 2013; Goodwin, Forman, Herbert, Butryn, & Ledley, 2011); and, eight were randomised controlled trials (RCT) (Brown, Whittingham, Boyd, McKinlay, & Sofronoff, 2014; Gregg, Callaghan, Hayes, & Glenn-Lawson, 2007; Hawkes, Pakenham, Chambers, Patrao, & Courneya, 2014; Hawkes et al., 2013; Lundgren, Dahl, Melin, & Kies, 2006; Lundgren, Dahl, Yardi, & Melin, 2008; Nordin & Rorsman, 2012; Rost, Wilson, Buchanan, Hildebrandt, & Mutch, 2012; Whittingham, Sanders, McKinlay, & Boyd, 2014). The studies using RCT and pre-post designs involved samples of people with: cancer (Feros et al., 2013; Hawkes et al., 2014; Hawkes et al., 2013; Rost et al., 2012), epilepsy (Lundgren et al., 2006; Lundgren et al., 2008), multiple sclerosis (Nordin & Rorsman, 2012), cardiac disease (Goodwin et al., 2011), type II diabetes (Gregg et al., 2007);and, paediatric cerebral palsy (Whittingham et al., 2014), brain injury (Brown et al., 2014), and life threatening illness (Burke et al., 2014). The RCT studies compared ACT to a waitlist control group (Whittingham et al., 2014), treatment as usual (TAU) (Brown et al., 2014; Hawkes et al., 2014; Hawkes et al., 2013), and other active treatments (including education, yoga, cognitive therapy, relaxation training, supportive therapy) (Gregg et al., 2007; Lundgren et al., 2006; Lundgren et al., 2008; Nordin & Rorsman, 2012; Rost et al., 2012; Whittingham et al., 2014). In studies using group-based analyses, one study had a sample size of 205 (Hawkes et al., 2014; Hawkes et al., 2013); however, most others had a small sample size (M = 23.00, SD = 13.03; range = 10 - 45) (Table 2.1 & 2.2).

Most (eight) of the included interventions were delivered at least in part within groups (Brown et al., 2014; Burke et al., 2014; Goodwin et al., 2011; Gregg et al., 2007; Lundgren et al., 2006; Lundgren et al., 2008; Nordin & Rorsman, 2012; Whittingham et al., 2014); with just three (Feros et al., 2013; Hawkes et al., 2014; Hawkes et al., 2013; Rost et al., 2012) delivered exclusively via one-to-one sessions with a therapist. The number of sessions ranged from 1-12. Most interventions were brief (*M* = 7 sessions, *SD* = 3.84), with six studies evaluating interventions of no more than 5 sessions (Burke et al., 2014; Goodwin et al., 2011; Gregg et al., 2007; Lundgren et al., 2006; Lundgren et al., 2008; Nordin & Rorsman, 2012). Participant drop-out from the included interventions was low with an average of 81.60% (*SD* 15.51; range 60 -100) of participants completing treatment. Three interventions showed 100% completion (Gregg et al., 2007; Lundgren et al., 2006; Lundgren et al., 2008) (Table 2.1.)

The case studies included a range of long-term conditions: HIV (Moitra et al., 2011; Skinta et al., 2014), diabetes (Nes et al., 2012), multiple sclerosis (Gillanders & Gillanders, 2014), stroke (Graham, Gillanders, et al., 2014) and sickle cell disease (Masuda et al., 2011). No included case study used an experimental design, one recorded reliable change (Gillanders & Gillanders, 2014), and one used session-by-session measurement of outcomes (Graham, Gillanders, et al., 2014) (Table 2.2).

Table 2.1. Summary of interventions, comparison groups and outcomes of included studies using pre- post- or RCT designs

Study	Design	POMRF Score (range 0 -40)	Disease Group	Primary Target of intervention	Format of intervention	No. of sessions (Total Hours)	N (% finished)	Control (brief descriotion)	Outcomes (Measures)	Improvement post intervention (Mean ES)	Improvement compared to control (Mean ES)	Maintained at follow-up
Brown et al., (2014)	RCT	24	Paediatric brain injury	Parenting intervention	Group & individual therapy	11 (17.5)	30 (85%)	TAU (access to rehab. Services)	Child behaviour & emotional problems (distress) (ECBI; SDQ)	(*d* = 0.84)	3/3 significant (*d* = 0.67)	2/3 maintained
									Dysfunctional parenting style (PS)	(*d* = 0.61)	2/2 significant (*d* = 0.65)	2/2 maintained
Burke et al., (2014)	Pre-post	10	Paediatric life threatening illness	Parental distress	Group therapy	5 (7.5)	11 (73%)	-	Parental distress (PCL-C; PECI)	5/5 significant (*d* = 1.12)	-	5/5 maintained
									Psychological flexibility (PPF)	2/3 significant (*d* = 0.72)	-	3/3 maintained
									Mindfulness (MAAS)	1/1 significant (*d* = 0.54)	-	1/1 maintained
Feros et al., (2013)	Pre-post	15	Cancer	QoL and distress	Individual therapy	9 (6.75)	45 (62%)	-	Distress (DT; DASS)	2/2 significant (*d* = 1.05)	-	2/2 maintained
									QoL (FACT)	1/1 significant (*d* = 0.56)	-	1/1 maintained
									Psychological flexibility (AAQ II)	1/1 significant (*d* = 0.64)	-	1/1 maintained
Goodwin et al., (2011)	Pre-post	15	Cardiac disease	Improving lifestyle	Group therapy	4 (6)	16 (75%)	-	Self-report diet (ASA-24)	3/3 significant (*d* = 1.27)	-	-
									Weight (BMI; lbs)	1/2 significant (*d* = 0.11)	-	-
									Self-report exercise (IPAQ)	0/1 significant (*d* = 0.54)	-	-
									Mindfulness (PHLMS)	1/2 significant (*d* = 0.31)	-	-
									Defusion (DDS)	0/1 significant (*d* = 0.23)	-	-
									Values (VGCM)	1/1 significant (*d* = 0.33)	-	-
									Psych. flexibility (FAAQ; PA-AAQ)	1/2 significant (d = 0.57)		

Study	Design	POMRF Score (range 0 -40)	Disease Group	Primary Target of intervention	Format of intervention	No. of sessions (Total Hours)	N (% finished)	Control	Outcomes (Measures)	Improvement post intervention (Mean ES)	Improvement compared to control (Mean ES)	Maintained at follow-up
Gregg et al., (2007)	RCT	21	Type II Diabetes	Improving diabetes self-management	Group therapy	1 (4)	43 (100%)	Education (7hr workshop)	No. in glucose control	1/1 significant -	1/1 significant (*d* = 0.61)	-
									HbA1c	1/1 significant (*d* = 0.42)	0/1 significant (*d* = 0.35)	-
									Self-management (DSCAM)	1/1 significant (*d* = 1.06)	1/1 significant (*d* = 0.68)	-
									Psychological flexibility (AADQ)	1/1 significant (*d* = 0.49)	1/1 significant (*d* = 0.78)	-
									Understanding (DCP)	1/1 significant (*d* = 0.37)	0/1 significant (*d* = 0.30)	-
Hawkes et al. (2013; 2014)*	RCT	30	Colorectal cancer	Improving lifestyle	Individual therapy	11 (?)	205 (72%)	TAU (access to educational materials)	Self-report physical activity (GLTEQ;)	- (*d* = 0.14)	0/3 significant (*d* = 0.06)	2/3 better than control at follow-up
									Self-report weight (BMI)	1/1 significant (*d* = 0.37)	1/1 significant (*d* = 0.23)	1/1 maintained
									Self-report diet (CCVFFQ)	4/6 significant -	3/6 significant (*d* = 0.20)	2/6 maintained
									Distress (BSI-18)	1/1 significant -	0/1 significant (*d* = 0.01)	0/1 maintained
									QoL (SF-36; FACT-C)	3/3 significant -	0/3 significant (*d* = 0.08)	0/3 maintained
									Psychological flexibility (AAQ II)	1/1 significant -	1/1 significant (*d* = 0.15)	0/1 maintained
									Mindfulness (MAAS)	1/1 significant -	0/1 significant (*d* = -0.01)	0/1 maintained

Study	Design	POMRF Score (range 0 -40)	Disease Group	Primary Target of intervention	Format of intervention	No. of sessions (Total Hours)	N (% finished)	Control	Outcomes (Measures)	Improvement post intervention (Mean ES)	Improvement compared to control (Mean ES)	Maintained at follow-up
Lundgren et al., (2006)	RCT	22	Epilepsy	Improve seizure control & QoL	Group & individual	4 (11)	14 (100%)	Supportive therapy (11 hrs)	Seizure intensity (SI)	(*d* = 1.21)	1/1 significant (*d* = 1.45)	1/1 maintained
									QoL (WHOQOL-BREF)	(*d* = 0.62)	0/1 significant (*d* = 0.37)	1/1 improved to follow-up
									Life satisfaction (SWLS)	(*d* = 0.73)	1/1 significant (*d* = 1.72)	1/1 maintained
Lundgren et al., (2008)	RCT	21	Epilepsy	Improve seizure control & QoL	Group & individual	4 (12)	10 (100%)	Yoga (12hrs)	Seizure intensity (SI)	1/1 significant (*d* = 1.3)	1/1 significant (*d* =1.14)	-
									QoL (WHOQOL-BREF)	1/1 significant (*d* = 0.81)	0/1 in favour of yoga (*d* = -0.38)	1/1 remained non-significant
									Life satisfaction (SWLS)	0/1 significant (*d* = 0.55)	0/1 significant (*d* = 0.12)	1/1 remained non-significant
Nordin & Rorsman (2012)	RCT	20	Multiple sclerosis	Treatment of distress	Group therapy	5 (?)	11 (?)	Relaxation training	Distress (HADS; BDI)	1/3 significant	0/3 significant	Remained non-significant
									Psychological flexibility (AAQ)	1/1 significant	0/1 significant	1/1 remained non-significant

Study	Design	POMRF Score (range 0 -40)	Disease Group	Primary Target of intervention	Format of intervention	No. of sessions (Total Hours)	N (% finished)	Control	Outcomes (Measures)	Improvement post intervention (Mean ES)	Improvement compared to control (Mean ES)	Maintained at follow-up
Rost et al., (2011)	RCT	17	Late-stage ovarian cancer	Treatment of distress	Individual therapy	12 (12)	25 (60%)	Cognitive therapy (12hrs)	QoL (FACT)	1/1 significant (*d* = 1.59)	1/1 significant (*d* = 1.35)	-
									Distress (POMS; BAI; BDI II)	3/3 significant (*d* = 1.87)	3/3 significant (*d* = 1.28)	-
									Acceptance coping (COPE)	1/1 significant (*d* = 1.58)	1/1 significant (*d* = 2.02)	-
									Mental disengagement (COPE)	1/1 significant (*d* = - 1.83)	1/1 significant (*d* = -3.49)	-
									Emotional control (CECS)	1/1 significant (*d* = -3.76)	1/1 significant (*d* = -6.11)	-
									Thought suppression (WBSI)	1/1 significant (*d* = -1.97)	1/1 significant (*d* = -3.02)	-
Whittingham et al., (2014)	RCT	23	Paediatric cerebral palsy	Parenting intervention	Group & individual	11 (17.5)	23 (91%)	1. WL control 2. Parenting intervention (13.5hrs)	Child behaviour & emotional problems (ECBI; SDQ)	(*d* = 0.25)	1.3/8 significant (*d* = 0.48) 2. 1/8 significant + 1 in favour of SSTP (*d* = 0.14)	2. 1/8 maintained
									Dysfunctional parenting style (PS)	(*d* = 0.77)	1.2/3 significant (*d* = 0.82) 2. 0/3 significant (*d* = 0.39)	2. 2/3 Improved to follow-up

Table 2.1. Footnotes.
AADQ = Acceptance and Action Diabetes Questionnaire; AAQ II = Acceptance and Avoidance Questionnaire II; ASA-24 = Automated Self-Administered 24-hr Dietary Recall; BAI = Beck Anxiety Inventory; BDI = Beck Depression Inventory; BDI II = Beck Depression Inventory II; BMI= Body Mass Index; BSI-18= Brief Symptom Inventory (18 item); CCVFFQ = Cancer Council Victoria Food Frequency Questionnaire; CECS = Courtland Emotional Control Scale; COPE = The COPE Questionnaire; DASS = Depression Anxiety and Stress Scale; DCP = Diabetes Care Profile; DSCAM = Diabetes Self Care Assessment Measure; DT = Distress Thermometer; ECBI = Eyberg Child Behavior Inventory; FAAQ = Food Acceptance and Action Questionnaire; FACITFS = Functional Assessment of Chronic Illness Therapy Fatigue Scale; FACIT-Sp = Functional Assessment of Chronic Illness Spiritual Well-being; FACT = Functional Assessment of Cancer Therapy; GLTEQ = Godin Leisure-Time Exercise Questionnaire; HADS = Hospital Anxiety and Depression Questionnaire; IPAQ = International Physical Activity Questionnaire; MAAS = Mindfulness Attention Awareness Scale; PA-AAQ = Physical Activity Acceptance and Action Questionnaire; PCL-C = PTSD Checklist-Civilian Version; PECI = Parent Experience of Child Illness; PGI = Post-traumatic Growth Inventory; PHLMS = Philadelphia Mindfulness Scale; POMS = Profile of Mood States; PPF = Parental Psychological Flexibility Questionnaire; SDQ = Strengths and Difficulties Questionnaire; SF-36 = Short-Form 36; SI = Seizure Index; SWLS = Satisfaction With Life Scale; VGCM = Values and Goals Clarity Measure; WBSI = White Bear Suppression Inventory; WHOQOL-BREF = World Health Organisation Quality of Life

*Large number of outcome variables; only those which appeared to be primary and secondary outcomes reported (post-traumatic growth, spirituality and fatigue excluded)

Table 2.2. Brief description of the case studies/series included in this review

Study	Population	N	Target of the intervention	No. of sessions	Intervention components	Changes in measured outcomes
Gillanders & Gillanders (2014)	Multiple Sclerosis	1	Distress and coping with trauma memories	10 individual and with partner	Couples and individual work involving: workability analysis; defusion; willingness; acceptance values elicitation; committed action. Mindfulness exercises also used.	Clinically significant improvements in depression and anxiety (HADs) – considered 'recovered' at the end of treatment. Improvement in psychological flexibility also evidenced.
Graham, Gillanders et al., (2015)	Stroke	1	Anxiety, stress and medically unexplained chest pain	9 individual sessions	Mindfulness (contact with the present moment; self as context); workability analysis; defusion; willingness and acceptance	Substantial improvements in anxiety and stress across the course of treatment. Smaller improvement in depression. Substantial improvement in psychological flexibility.
Masuda et al., (2011)	Sickle cell disease	1	Improving functioning and QoL	8 individual sessions with child and parents	Family sessions focussing on perspective modification, values clarification, acceptance, mindfulness and committed action	Improvement to follow-up in child's self-rated and parent rated functioning. Improvement in average pain levels. Improvement in QoL at follow-up. Improvement in psychological flexibility and in parent's acceptance of child's illness.
Moitra et al., (2011)	HIV	16	Improving adherence to HIV medication	3-5 group sessions	Exploring willingness to accept HIV diagnosis, fusion with HIV fears, experiential avoidance of HIV self-management. Mindfulness, values and committed action explored in relation to adherence.	Trends towards improvement in viral load. The majority of participants reported that the intervention was helpful.
Nes et al., (2012)	Diabetes	11	Improving diabetes self-management via a smartphone-delivered intervention	Unclear	Identifying values and valued activities in the context of diabetes self-management.	A small change in HBA1c was noted as well as improvements in QoL, diabetes-related distress and BMI. Generally patients reported being satisfied with the intervention.
Skinta et al., (2014)	HIV	3	Reducing HIV self-stigma	8 group-based sessions	Stigma and experiential avoidance; valued-living and committed action; acceptance and mindfulness (self-as-context); willingness.	Improvements in HIV self-stigma which were maintained at two month follow-up. Some evidence of improvement in psychological flexibility

HADS=Hospital Anxiety and Depression Scale; QoL = Quality of Life

Table 2.3. Between-group standardised and unstandardised effect sizes (with 95% confidence interval), for each variable.

Study	Outcomes (Measures)	Improvement compared to control – ES and (95%CI) for each measure*
Brown et al., (2014)	Child behaviour & emotional problems (distress) (ECBI; SDQ)	3/3 significant (mean *d* = 0.67) -ECBI intensity 0.90 (0.34, 1.46) -ECBI problem 0.76 (0.20, 1.31) -SDQ 0.50 (-0.04, 1.05)
	Dysfunctional parenting style (PS)	2/2 significant (mean *d* = 0.65) - Laxness 0.76 (0.21, 1.32) - Over-reactivity 0.54 (-0.01, 1.08)
Gregg et al., (2007)	No. in glucose control	1/1 significant 0.61*
	HbA1c	0/1 significant 0.42 (-0.03, 0.85)†
	Self-management (DSCAM)	1/1 significant 0.22 (-0.22, 0.65)†
	Psychological flexibility (AADQ)	1/1 significant 0.38 (-.06, 0.82)†
	Understanding (DCP)	0/1 significant 0.45 (0.01, 0.89) †
Hawkes et al. (2013; 2014)*	Self-report physical activity (GLTEQ;)	0/3 significant (*d* = 0.06) - moderate 0.10 (-0.11, 0.32); 16.5 mins (-7.4, 40.5) - vigorous - 0.05 (-0.26, 0.16); -2.7 mins (-14.2, 8.9) - mod-vig 0.11 (-0.10, 0.31); 11.5 mins (-18.82, 41.9)
	Self-report weight (BMI)	1/1 significant *d* = 0.23 (0.02, 0.44); -0.5 BMI (-1.0, 0.0)
	Self-report diet (CCVFFQ)	3/6 significant (*d* = 0.20) - Fat 0.37 (0.16, 0.59); % fat -8.5 (-13.4, 3.6) - Sat fat 0.33 (0.12, 0.55); % sat fat -3.5 (-5.7, 1.2) - Fibre -0.10 (-0.31, 0.11); g/day -0.7 (-2.2, 0.8) - Fruit 0.15 (-0.06, 0.36); servings 0.2 (-0.0, 0.4) - Veg 0.30 (0.09, 0.52); servings 0.4 (0.2, 0.7) - Alcohol 0.12 (-0.09, 0.33); units -1.4 (-3.7, 1.0)
	Distress (BSI-18)	0/1 significant *d* = 0.01 (-0.20, 0.22)
	QoL (SF-36; FACT-C)	0/3 significant (*d* = 0.08) -PCS 0.0 (-0.21, 0.21) -MCS 0.09 (-0.12, 0.30) -FACT-C QoL 0.15 (-0.06, 0.36)
	Psychological flexibility (AAQ II)	1/1 significant *d* = 0.15 (-0.06, 0.36)
	Mindfulness (MAAS)	0/1 significant *d* = 0.00-
Lundgren et al., (2006)	Seizure index (SI)	1/1 significant† 1.89 (0.99, 2.80)
	QoL (WHOQOL-BREF)	0/1 significant *d* = 0.37 (-0.40, 1.13)
	Life satisfaction (SWLS)	1/1 significant (*d* = 1.72)* 1.77 (0.89, 2.67)
Lundgren et al., (2008)	Seizure index (SI)	1/1 significant *d* =1.14 (0.14, 2.14)
	QoL (WHOQOL-BREF)	0/1 in favour of yoga *d* = -0.38 (-1.32, 0.56)
	Life satisfaction (SWLS)	0/1 significant *d* = 0.12 (-0.81, 1.05)
Nordin & Rorsman (2012)	Distress (HADS; BDI)	0/3 significant*
	Psychological flexibility (AAQ)	0/1 significant
Rost et al., (2011)	QoL (FACT)	1/1 significant *d* = 1.01(0.27, 1.77) †
	Distress (POMS; BAI; BDI II)	3/3 significant† (*d* = 1.28)*

		Distress 1.34 (0.56, 2.12) Depression 0.86 (0.12, 1.59) Anxiety 1.20 (0.44, 1.97)
	Acceptance coping (COPE)	1/1 significant* 3.17 (2.12, 4.23)
	Mental disengagement (COPE)	1/1 significant* 9.90 (7.34, 12.46)
	Emotional control (CECS)	1/1 significant* 4.38 (3.08, 5.68)
	Thought suppression (WBSI)	1/1 significant* 1.94 (1.08, 2.79)
Whittingham et al., (2014)	Child behaviour & emotional problems (ECBI; SDQ)	1.3/8 significant (*d* = 0.48) -ECBI Intensity 0.77 (0.17, 1.38) -ECBI Problem 1.34 (0.68, 1.98) -SDQ Emot sympts 0.37 (-0.22, 0.96) -SDQ Conduct probs 0.25 (-0.34, 0.84) -SDQ Hyperactivity 0.22 (-0.37, 0.80) -SDQ Peer Probs 0.47 (-0.12, 1.07) -SDQ Prosocial 0.10 (-0.49, 0.68) -SDQ Impact 0.30 (-0.29, 0.89) 2. 1/8 significant + 1 in favour of SSTP (*d* = 0.14) -ECBI Intensity 0.43 (-0.16, 1.02) -ECBI Problem 0.58 (-0.02, 1.17) -SDQ Emot sympts -0.41 (-1.0, 0.18) -SDQ Conduct probs -0.18 (-0.76, 0.41) -SDQ Hyperactivity 0.11 (-0.47, 0.70) -SDQ Peer Probs-0.32 (-0.91, 0.26) -SDQ Prosocial 0.29 (-0.30, 0.88) -SDQ Impact 0.65 (0.06, 1.26)
	Dysfunctional parenting style (PS)	1.2/3 significant (*d* = 0.82) -PS Laxness 0.43 (-0.16, 1.03) -PS Over reactivity 1.11 (0.48, 1.76) -PS Verbosity 0.93 (0.31, 1.54) 2. 0/3 significant (*d* = 0.39) -PS Laxness 0.06 (-0.53, 0.64) -PS Over reactivity 0.66 (0.06, 1.26) -PS Verbosity 0.44 (-0.15, 1.04)

†ES calculation differs from that published in paper, to enable calculation of 95% CI

*unable to calculate ES and/or 95% CI from the information provided in the publication

Quality of the studies using group-based statistics

Study quality (excluding case studies) was generally quite low (*M* = 19.82, *SD* = 5.38; range = 10-30) with just 6 of the 11 (Brown et al., 2014; Gregg et al., 2007;

Hawkes et al., 2014; Hawkes et al., 2013; Lundgren et al., 2006; Lundgren et al., 2008; Whittingham et al., 2014) studies receiving more than half of the available points on the POMRF. We used this cut-off (>20 points on the POMRF) to denote a higher-quality study. The highest quality study (Hawkes et al., 2014; Hawkes et al., 2013) achieved a score of 30 (Table 2.2 and Appendix vi).

Several consistent strengths were apparent across studies. For example, all studies showed a fair description of statistical methods and presentation of results, with all but one (Nordin & Rorsman, 2012) achieving a maximum score. All studies gave at least a fair description of the intervention and/or were able to direct readers to an intervention manual, with 6 of 11 (Brown et al., 2014; Gregg et al., 2007; Lundgren et al., 2006; Lundgren et al., 2008; Nordin & Rorsman, 2012; Whittingham, 2014) achieving a maximum score. All studies used outcomes measures that were psychometrically adequate (specific and/or reliable), and all included a population which appeared adequately representative of a clinical sample.

Nonetheless, only one study used trial evaluators who were blinded to the treatment condition (Hawkes et al., 2014; Hawkes et al., 2013). Also, just one (Lundgren et al., 2006) showed a clear effort to control for concomitant treatments. Studies often introduced a systematic condition/therapist confound by having one therapist per condition or did not report the number of therapists (Burke et al., 2014; Feros et al., 2013; Lundgren et al., 2008; Rost et al., 2012). Just three studies (Gregg et al., 2007; Hawkes et al., 2014; Hawkes et al., 2013; Whittingham et al., 2014) included an a priori power/sample size calculation; while consideration of clinical significance was apparent in just four studies (Feros et al., 2013; Gregg et al., 2007; Hawkes et al., 2014; Hawkes et al., 2013; Whittingham et al., 2014). Where control interventions were included these were frequently unequal in duration to the ACT intervention (evident in 4 of 6 RCTs) (Brown et al., 2014; Gregg et al., 2007; Hawkes et al., 2014; Hawkes et

al., 2013; Whittingham et al., 2014). Long-term (i.e. 12 month) follow-up was evident in just three studies (Hawkes et al., 2014; Hawkes et al., 2013; Lundgren et al., 2006; Lundgren et al., 2008).

Applications and their emerging efficacy

The returned ACT interventions sought to engender change in a range of outcomes. An analysis of the emerging efficacy for ACT in each outcome is described below.

Distress

Five studies evaluated whether ACT interventions can reduce distress in people with long-term conditions (Burke et al., 2014; Feros et al., 2013; Hawkes et al., 2014; Hawkes et al., 2013; Nordin & Rorsman, 2012; Rost et al., 2012). With one exception (Nordin & Rorsman, 2012), all observed a significant improvement in most measures of distress following ACT. Where reported, this change showed a large effect size (range d = 1.05 – 1.86) (Burke et al., 2014; Feros et al., 2013; Rost et al., 2012).

Three of these studies used RCT designs (Hawkes et al., 2014; Hawkes et al., 2013; Nordin & Rorsman, 2012; Rost et al., 2012). The highest quality study compared a telephone-delivered ACT intervention for people with colorectal cancer to TAU (Hawkes et al., 2014; Hawkes et al., 2013). Here, no significantly greater improvement was observed in the ACT group compared to TAU at post-intervention (d =-0.01) or follow-up. However, distress could be considered a secondary outcome in this intervention which primarily sought to improve lifestyle (diet and weight). Accordingly, participants were not included based on a high level of distress, making it less likely that a post-intervention improvement in this outcome might be detected.

Two lower-quality studies, with interventions more explicitly targeting distress, compared ACT to active treatments (relaxation training (Nordin & Rorsman, 2012)] and

cognitive therapy (Rost et al., 2012)]). Rost et al., (2012) evaluated a 12-session ACT treatment, delivered in a one-to-one format, for women with late-stage ovarian cancer. A significantly greater reduction occurred in all measures of distress following ACT when compared to cognitive therapy, and a very large average effect size was observed for this comparison (d = 1.28); however, the 95% confidence intervals surrounding the constituent effect sizes were quite broad, and also included very trivial effect sizes (see Table 2.3). This suggests little precision in this estimate, and that the 'true' effect size might be much smaller than this mean value. Nordin & Rorsman, (2012) evaluated a brief group-delivered ACT intervention for distress in multiple sclerosis. There was no significant benefit of ACT over relaxation training post-intervention or at follow-up. However, this trial appeared severely underpowered, with just 11 participants beginning the ACT condition.

Two case studies detail ACT applied to distress in neurological illnesses (Gillanders & Gillanders, 2014; Graham, Gillanders, et al., 2014). Gillanders & Gillanders (2014) describe an intervention involving an individual with multiple sclerosis and their partner; sessions were aimed at adjustment to progressing multiple sclerosis, against a background of childhood trauma. The case study showed clinically significant improvements in psychological flexibility and distress. Graham, Gillanders et al., (2015) outline the application of an ACT intervention to post-stroke anxiety – detailing how acceptance, workability analysis and present-moment-awareness can be used to manage illness-related fears. Subsequent improvements in stress and post-stroke anxiety were noted in this study.

Summary

With one exception (Nordin & Rorsman, 2012), ACT interventions were consistently associated with post-intervention improvements in distress. However, bar

one highly-supportive but lower-quality study (Rost et al., 2012), there is little evidence to suggest that ACT is superior to TAU or other psychological interventions, or that the consistent post-intervention improvements observed are little more than regression to the mean, placebo or the result of non-specific effects of therapy.

Parenting of children with long-term conditions

Two higher quality studies evaluated ACT for improving the parenting, and subsequent emotional and behavioural problems, of children with brain injury (Brown et al., 2014) and cerebral palsy (Whittingham et al., 2014) respectively. Both studies included an ACT-enhanced version of an established parenting program (Stepping Stones Triple P (Sanders, 2012)]), and had similar sample sizes (N = 30 (Brown et al., 2014)] and N = 23 (Whittingham et al., 2014)]). Following the intervention both observed a moderate (d = 0.61 – d = 0.77) improvement in parenting. With one (Brown et al., 2014) noting a large and statistically significant (d = 0.84) subsequent improvement in child behavioural and emotional problems; while the other observed a small change (d = 0.25) in this outcome. When compared to waitlist control or TAU, both observed significant improvements of moderate-to large size (d = 0.65 – 0.82) in dysfunctional parenting styles post intervention; with most measures showing statistically significant improvements; withthe 95% confidence intervals including small to large effect sizes . However, while a subsequent impact on child behavioural and emotional problems in favour of ACT was evident in these comparisons, they showed slightly smaller effect sizes (d = 0.48 – 0.67), with 95% confidence intervals ranging from negative values to large effect sizes depending on the domain (Table 2.3).

Whittingham et al., (2014), included a further trial arm: comparing the ACT enhanced Stepping Stones Triple P intervention to the Stepping Stones Triple P intervention alone. Here, just 1 of 8 child behavioural and emotional problems variables

showed greater response to ACT, with one showing a significantly greater response to Stepping Stones Triple P alone. However, on average, a small effect size (d = 0.14) in favour of ACT was noted. No post intervention between-group differences in dysfunctional parenting were apparent, but at six-month follow-up the ACT group had significantly better outcomes on 2 of 3 measures. However, it was notable that the ACT-embedded intervention was 4 hours longer than the Stepping Stones Triple P alone. Therefore, these differential outcomes may be related to differences in intensity/dosage.

Masuda et al., (2011) present a detailed case study of a family-based ACT intervention for improving functioning in a teenager with sickle cell disease. A range of ACT techniques were applied: perspective-taking, experiential acceptance, mindfulness and values-clarification in relation to parenting. Post-intervention improvements were noted in child- and parent-reported outcomes (such as functioning and parental acceptance of their child's illness) at the end of the intervention, with further improvement to three-month follow-up.

Summary

Two higher-quality studies support the application of ACT interventions to improving the parenting of children with long-term conditions. ACT showed effects which were greater than TAU for improving dysfunctional parenting. Further, results from one high-quality study (Whittingham et al., 2014) suggest that incorporating ACT into an established parenting intervention may significantly increase its efficacy for parenting and possibly children's emotional regulation or behaviour. Therefore ACT components appeared to add value to an established parenting treatment. However, a caveat is that the ACT-embedded intervention was slightly longer in duration than the established parenting intervention alone, and differential outcomes may be explained by this; thus, better controlled studies are required.

Self-management/lifestyle

Three studies evaluated ACT for improving disease self-management and/or lifestyle (Goodwin et al., 2011; Gregg et al., 2007; Hawkes et al., 2014; Hawkes et al., 2013). In a lower-quality study Goodwin et al., (2012) evaluated a brief group-based ACT intervention for improving lifestyle in people with cardiac diseases. This pre-post design observed significant and large improvements in all aspects of self-reported diet (3 of 3 measures significant; d = 1.27), and small, less consistent improvements in weight (1 of 2 measures significant; d = .11). While there was no significant improvement in self-reported exercise (0 of 1 measures significant), a moderate effect size was observed for this comparison (d = 0.54).

In a higher-quality study, Hawkes et al., (2013, 2014) assessed a more intensive intervention (individual sessions, more sessions) for improving lifestyle in colorectal cancer survivors. Similarly, significant improvements were seen in self-reported weight (1 of 1 measures significant) and most aspects of diet (4 of 6 measures significant). Compared to a TAU condition in which educational materials regarding methods to reduce cancer risk were made available, significant improvements were observed in self-reported weight and diet, and these were mostly maintained at 12 month follow-up. However, this study had a very large sample size (N = 205) and while most were statistically significant, these changes showed a small effect size (d = 0.20 – 0.23), with most 95% confidence intervals also lying within the small effect size range (Table 2.3). No improvements in self-rated physical activity over TAU were apparent at post-intervention (0 of 3 measures significant, d =0.06); however, by 12 month follow-up the ACT group were significantly more active (2 of 3 measures significant).

In another high-quality study, Gregg et al., (2007) evaluated an ACT-based diabetes self-management workshop. Compared to a diabetes education group, they observed a significant improvement of moderate size in self-management (d = .68), and

subsequently, a significantly greater proportion were in objectively measured glucose control (*d* = 0.61). However, the 95% confidence intervals suggest little precision in the effect size estimates (Table 2.3), and no significant difference between groups in mean HbA1c blood levels was apparent (*d* = 0.35).

One case series describes a smartphone-based intervention for improving self-management in diabetes (Nes et al., 2012). The smartphones included diaries with written situational feedback, alongside face-to-face and telephone consultation with clinicians. The intervention was experienced as acceptable and few technical problems were encountered.

Summary

There is emerging evidence that ACT can improve disease self-management and life-style. One higher-quality study showed support when compared to TAU, though the size of this effect appeared very small (Hawkes et al., 2014; Hawkes et al., 2013) and many outcomes were self-reported. In another higher-quality study a significant improvement of moderate size was observed in biochemical measures of disease self-management. Thus, at present, a small number of studies suggest that ACT may be effective in these contexts. However, given that so few comparisons could be made (one comparison with TAU; one comparison with an active treatment) this evidence is very preliminary.

Quality of Life (QoL)

Five studies included QoL as an outcome measure (Feros et al., 2013; Hawkes et al., 2014; Hawkes et al., 2013; Lundgren et al., 2006; Lundgren et al., 2008; Rost et al., 2012). All showed significant improvements in QoL following ACT (Feros et al., 2013; Hawkes et al., 2014; Hawkes et al., 2013; Lundgren et al., 2008; Rost et al., 2012); with moderate-to-large effect sizes (d = 0.56 to d = 1.59), where these were reported or could

be calculated (Feros et al., 2013; Lundgren et al., 2006; Lundgren et al., 2008; Rost et al., 2012).

One higher quality study compared ACT to TAU for improving lifestyle and QoL in colorectal cancer survivors (Hawkes et al., 2014; Hawkes et al., 2013). Here no significant improvements over TAU were observed in QoL domains (Hawkes et al., 2014; Hawkes et al., 2013), with a corresponding small average effect size (d = 0.08), quite precise 95% confidence intervals suggesting that a small effect size is likely (Table 2.3).

Three studies compared ACT to active treatments (Lundgren et al., 2006; Lundgren et al., 2008; Rost et al., 2012). Two were higher-quality, albeit likely slightly underpowered, studies investigating the efficacy of the same ACT intervention for improving seizure control and QoL in epilepsy (Lundgren et al., 2006; Lundgren et al., 2008). When compared to supportive therapy, a small non-significant improvement in QoL was observed at post-intervention (d = 0.37, 95% CI -0.40, 1.13)(Lundgren et al., 2006), which became significant at one year follow-up (Lundgren et al., 2006). However, when compared to yoga, a small effect size was in favour of yoga was observed (d = -0.38, 95% CI -1.32, 0.56) (45). One lower-quality study investigated the impact of one-to-one ACT sessions on distress and QoL in women with late-stage ovarian cancer (Rost et al., 2012). When compared to an intervention of the same intensity which was reminiscent of cognitive therapy (involving cognitive restructuring, problem-solving, relaxation training), an average large (d = 1.28) significantly greater improvement in QoL was apparent in the ACT group; however, estimated 95% confidence intervals also included small effect sizes (Table 2.3).

Summary

The existing evidence presents an inconsistent picture of the efficacy of ACT for improving QoL in long-term conditions. While a consistent post-intervention

improvement in QoL was apparent, it is unclear whether ACT interventions are more effective than TAU or other active treatments, or again if post-intervention improvements are the result of placebo or non-specific therapy factors.

Psychological Flexibility

Six studies assessed post-ACT intervention changes in psychological flexibility, as measured with the Acceptance and Avoidance Questionnaires (Burke et al., 2014; Feros et al., 2013; Goodwin et al., 2011; Gregg et al., 2007; Hawkes et al., 2014; Hawkes et al., 2013; Nordin & Rorsman, 2012). All observed significant pre-to-post intervention improvements following ACT in at least half included measures of psychological flexibility. Where it was reported or could be calculated (Burke et al., 2014; Feros et al., 2013; Goodwin et al., 2011; Gregg et al., 2007), this change showed a small-to-moderate effect size (d =0.57-0.72).

One study compared changes in psychological flexibility to TAU. This higher-quality trial of ACT for people with colorectal cancer observed a comparatively greater improvement in psychological flexibility in the experimental group (Hawkes et al., 2014; Hawkes et al., 2013). However, this difference had a very small effect size (d = 0.15, 95% CI -0.06, 0.36) and it was not maintained at 12 month follow-up.

Two studies which compared an ACT intervention to active treatments returned divergent results. In a higher-quality study, Gregg et al., (2007) evaluated an ACT self-management workshop for diabetes self-regulation, compared to an educational diabetes self-management workshop alone. Post-intervention changes in psychological flexibility were greater in the ACT group, with a moderate effect size (d = 0.78); However, the estimated 95% CI of this effect size was very broad, also including trivial effect sizes (Table 2.3). A lower-quality evaluation of an ACT intervention for people with multiple sclerosis observed no significant improvement compared to relaxation training, since

both interventions showed significant improvement by post intervention (Nordin & Rorsman, 2012).

Summary

Improvements in psychological flexibility following ACT were evident across studies. One higher- quality study reported this improvement to be greater than TAU, albeit with a very small effect size (Hawkes et al., 2014; Hawkes et al., 2013). Another observed greater improvement following ACT when compared to education (Gregg et al., 2007); however, a lower-quality study found no evidence of this effect when compared to relaxation training, since improvements was noted in both interventions (Nordin & Rorsman, 2012). Thus, while there are few studies, the evidence to date suggests that psychological flexibility improvement following ACT may be slightly greater than for treatment as usual and there is little to suggest that it is significantly less effective than other treatments.

Symptom Control

Two higher-quality studies evaluated the efficacy of ACT for directly controlling symptoms (Lundgren et al., 2006; Lundgren et al., 2008). Both included the same ACT intervention, which comprised sessions teaching behavioural methods for improving seizure control (Lundgren et al., 2006; Lundgren et al., 2008). These observed significantly greater improvements in seizure severity following ACT, when compared to supportive therapy (Lundgren et al., 2006) or yoga (Lundgren et al., 2008); with large effect sizes observed for these comparisons (d =1.4-1.45), and estimated 95% confidence intervals all lying within the large range (Table 2.3). However, both studies were undertaken by the same research group; thus replications by other groups are required. It could also be argued that the behavioural seizure control methods were the active component here and not ACT.

Summary

With two supportive higher quality studies the evidence supporting the application of ACT to seizure control is promising. However, further independent replication is required, where the effects of ACT-enhanced seizure control methods can be disentangled from those of seizure control methods alone.

Adherence

No studies using pre-post- or RCT designs evaluated ACT for improving adherence to medication. However, two case series were returned. These demonstrated a smartphone-delivered intervention to prompt self-management behaviours (including medication adherence) in diabetes (Nes et al., 2012), and the use of acceptance, values and committed action exercises to improve adherence to HIV medication (Moitra et al., 2011). Both interventions were judged as acceptable by participants and post-intervention trends towards improvements in HIV biomarkers, HBA1C and fasting blood glucose were observed.

Summary

Two case studies provide example applications of ACT in this context, with encouraging changes in outcomes. Therefore, larger-scale evaluations of ACT for improving adherence are warranted.

Stigma

One case study described an ACT-based group for reducing HIV- related self-stigma (Skinta et al., 2014). This emphasised the defusion, self-as-context, and values aspects of ACT, and some participants showed an improvement in self-stigma across sessions.

Summary

This further application demonstrates the versatility of the ACT model. However, the design enabled no comment on emerging efficacy.

DISCUSSION

A summary of ACT's use in Long-term conditions

ACT has been applied across many long-term conditions, for example: cancer (Feros et al., 2013; Hawkes et al., 2014; Hawkes et al., 2013; Rost et al., 2012), epilepsy (Lundgren et al., 2006; Lundgren et al., 2008), paediatric illness (Brown et al., 2014; Burke et al., 2014; Whittingham et al., 2014), cardiac disease (Goodwin et al., 2011), multiple sclerosis (Nordin & Rorsman, 2012), and diabetes (Gregg et al., 2007). Here ACT has been used to elicit change in a range of outcomes, from improving lifestyle/disease self-management (Gregg et al., 2007; Hawkes et al., 2014; Hawkes et al., 2013), and symptom control (Lundgren et al., 2006; Lundgren et al., 2008) to reducing distress (Nordin & Rorsman, 2012; Rost et al., 2012) and improving QoL (Feros et al., 2013; Lundgren et al., 2006; Lundgren et al., 2008). Several case studies gave detailed description of the process of applying ACT to distinct clinical problems, including: family intervention for functional impairment in an adolescent with sickle cell disease (Masuda et al., 2011); couples work for trauma and distress in MS (Gillanders & Gillanders, 2014); one-to-one sessions for post stroke anxiety (Graham, Gillanders, et al., 2014); a smart-phone delivered diabetes self-management intervention (Nes et al., 2012); and, interventions to improve self-stigma (Skinta et al., 2014) and non-adherence in HIV (Moitra et al., 2011).

The range of long-term conditions and applications demonstrates the flexibility of the ACT model and also reflects the extent to which practitioners (Thewes et al., 2014) and clinical researchers (Angiola & Bowen, 2013; Graham, Simmons, et al., 2014; Hadlandsmyth et al., 2013; Kangas & McDonald, 2011; Karekla & Constantinou, 2010; Moitra et al., 2011) working with long-term conditions have embraced ACT. Indeed, the number of intervention studies is increasing each year, with almost half the

included studies published in 2014 (the year preceding the systematic search). It is arguable as to whether the wide-spread adoption of ACT is recognition that it is particularly applicable to long-term conditions, or is a therapeutic fad. However, given that research in this area is young - the first included ACT intervention study was published in 2006 (Lundgren et al., 2006) - time is needed before this can be established.

It has been suggested that ACT might be usefully applied to medication non-adherence (Hadlandsmyth et al., 2013; Moitra et al., 2011). However, despite promising findings in case studies (Moitra et al., 2011; Nes et al., 2012), no comprehensive trials of this were evident in the present review. This is a missed opportunity for two reasons: 1) Non-adherence to medication is a major public health problem, with an estimated 30-50% of medication not taken as recommended, and subsequent serious repercussions (Horne et al., 2005); 2) adherence/non-adherence is a behaviour with a range of cognitive-behavioural correlates (Horne & Weinman, 1999); (Daley, Myint, Gray, & Deane). Therefore psychological intervention (potentially ACT) should be considered a first-line intervention (Petrie et al., 2012; Petrie & Weinman, 2012).

State of the evidence

It is a consistent finding that ACT is associated with improved outcomes across applications within long-term conditions. However, the paucity of studies using RCT designs (per application) and the general low quality of studies, meant it was unclear whether this was due to the intervention, non-specific therapy factors, placebo effect or regression to the mean. Indeed, many studies had very small sample. This is problematic because a negative correlation between sample size and effect sizes exists (Slavin & Smith, 2009) (Slavin, 2009). Critics believe this to be due to an interaction between the large variability of results in underpowered studies, and a publication bias.

Meaning that only those with statistically significant results, and thus very large effect sizes (given the small sample size) are published. The large proportion of such underpowered studies in this review leads one to wonder at the extent to which this phenomenon skews our results. Relatedly, the effect sizes of several very positive ACT studies (Rost et al., 2012; Gregg et al., 2006) had broad 95% confidence intervals, suggesting that the 'true' effect size could also lie in the smaller, or even the negative ranges. Thus, further higher quality studies are thus required to establish reliable/valid effect size estimates.

Thus, an overall comment on this emerging field of ACT applications is that, whilst findings to date are encouraging for some applications, much more high-quality research is needed before any application could be considered to have comprehensive empirical support (for example criteria see (Chambless & Hollon, 1998; Öst, 2008, 2014)) Nonetheless, there is emerging evidence that ACT may be effective in some contexts; With supportive higher-quality studies and little convincing counter-evidence, ACT shows promising application for improving the parenting of children with chronic illness, seizure control in epilepsy, psychological flexibility, and possibly self-management/lifestyle.

A further consideration regarding efficacy, is that the included interventions tended to have a very low number of sessions. A contemporaneous review of ACT interventions applied to anxiety (Swain et al., 2013) found that just 17% of ACT interventions were of 5 sessions or less compared to 45.5% in the present review, with many included interventions delivered by phone or in groups. The included interventions could therefore be considered a 'low-dosage' of psychotherapy. Since a dose-effect relationship has been noted in psychotherapy, with those receiving more sessions having better final outcomes (Kopta, 2003), one might expect a smaller impact of ACT within the current context than in mental health. Thus, researchers investigating

the efficacy of ACT interventions for long-term conditions should consider if they are providing interventions which are of sub-optimal intensity. Since long-term conditions are often disabling, life-threatening and can be accompanied by mood disturbance, one might reason that high-intensity interventions are required to elicit optimal changes.

Methodological Suggestions

The general low-quality of available evidence regarding efficacy is in contrast to the apparent clinical adoption of ACT for long-term conditions (Hadlandsmyth et al., 2013; Kangas & McDonald, 2011; Moitra et al., 2011; Thewes et al., 2014). This highlights the need for higher quality investigations of ACT in these contexts. Future studies should build on existing methodological strengths in: the presentation of results; description of treatment; the use of specific and reliable outcome measures; and the inclusion of representative samples. However, considerable improvement in methodology is required, with more RCTs needed. Below is a list of ways to improve trial methodology based on current limitations.

1.) Trained evaluators who are blind to condition allocation should assess study variables at each stage of the trial.

2.) Interventions should include at least two therapists per treatment; this is required to disentangle the effects of the intervention from the clinician delivering the intervention.

3.) A priori power calculations are required: several studies appeared underpowered and it was unclear to what extent.

4.) Concomitant treatments (other medications/interventions which might affect mood or behaviour) should be clearly described.

5.) Active control interventions should be clearly described and matched for length, intensity, components and clinician allegiance.

6.) Non-active control conditions should also be described clearly. In particular, detail should be given regarding the composition of TAU control conditions.

7.) Long-term follow-up is required (at least 12 months).

8.) To comment on impact, a priori indicators of clinical significance should also be used, and economic analysis considered.

9.) When behaviours are outcomes, direct measurement should be included (for example, pedometers to record physical activity, and biochemical measures of adherence) (Miller & Hays, 2000; Tudor-Locke & Myers, 2001).

10.) Finally, to aid the post-hoc calculation of effect-sizes, means and standard-deviations (or standard errors) for pre- and post-intervention measurements should be supplied. Further, to allow for alternative methods of calculating the effect size of within-group changes, correlation co-efficients between pre- and post- intervention variables could be provided as supplementary information (see Lakens, 2013 for a debate on this topic).

The design of ACT case study research could also be improved. Session-by-session measurement of outcomes and reliable change in outcomes were rarely reported. While case studies are arguably most useful as detailed descriptive accounts of the process of applying interventions, this does not mean that evaluation of change in outcomes is unimportant. Single case experimental designs can be used (Smith, 2012), and tools are available to assess the statistical significance of changes across the period of intervention (Borckardt et al., 2008; Morley, 2014), as well as mediation via cross-lagged correlation (Borckardt et al., 2008). Indeed, in rare illnesses, the highest level of evidence may be derived from such research.

Limitations

Several limitations are implicit in the present review. First, owing to the existence of few RCTs per application, a detailed meta-analyses was omitted from the present review. However, given the rapid increase in intervention studies with long-term conditions, this may soon be recommended.

A first limitation was the moderate level of inter-rater agreement in classifying the quality of the studies, which also indicates substantial disagreement between reviewers. This may have occurred for a number of reasons: it may reflect a lack of clarity/validity/reliability in the items of the quality assessment measure (POMRF), or indeed individual differences between raters (different training backgrounds, or individual differences in understanding of the texts/items). To enable more simple quantification and communication of the quality of studies in relation to their findings, a dichotomous cut-off for classifying studies as higher-quality and lower-quality was used. This division could be considered somewhat arbitrary, and we would thus encourage readers to also reflect on individual quality ratings of each study (Table 1; Appendix vi) in conjunction with the findings of the review. A recent review used z-scores (>1 = high quality; < -1 = low quality) to indicate high and low quality (Swain et al., 2013); however, given the low number of studies returned this was not possible here. Also, some items of the POMRF are of limited applicability to long-term conditions. Here two items regarding the certainty of diagnosis appeared superfluous and no assessment of whether outcomes were self-report or directly measured was included. Composition of specific quality assessment criteria for psychological intervention studies with long-term conditions is recommended.

Where effect-sizes were not already reported in RCTs we calculated effect size based on comparison between post-intervention scores. The accuracy of the resultant effect size thus relies on the assumption that pre-intervention scores are equivalent

between conditions–which seems unlikely. Also, when calculating within-group effect sizes we were unable to account for the correlation between the pre- and post-intervention variables; therefore, these effect sizes may be slightly inaccurate (see Lakens, 2013).

Finally, we did not include studies with chronic pain populations, which might be considered an oversight since chronic pain is often experienced in long-term conditions (Clifford & Trotter, 1984; Jensen et al., 2008). The exclusion was for two reasons: First and foremost, due to the implication that ACT is being used clinically but that there has been no systematic review of ACT specifically in long term conditions, this reviewed aimed to collate intervention studies accurately describe the field. In contrast to the literature described in the present review, ACT interventions for chronic pain are well-reviewed elsewhere (Veehof et al., 2011; McCracken & Vowles, 2014). Here, estimation of pooled effect sizes suggests that ACT is effective for chronic pain, with a small-to-medium effect size (Veehof et al, 2011). Secondly, interventions for chronic pain, such as those included in the preceeding reviews, often include people with pain which is not generated as part of a chronic disease/long-term condition (e.g. back pain as a result of aging or sports injury, period pain, or an unknown cause) (Veehof et al., 2011; Elliott et al., 1999). Indeed, here in the UK people with chronic pain are often treated by specialist pain management services (Barker & McCracken, 2013) , such services being somewhat distinct from services for chronic health conditions – e.g. Psycho-oncology, Neuropsychology etc.

CONCLUSION

ACT has been applied in many different ways within a range of long-term conditions. However, there have been no trials of ACT for improving medication non-adherence. Most of the included studies were low quality and there were very few RCTs.

Therefore, ACT interventions are not yet well established for use in long-term conditions. Nonetheless, encouraging results suggest that these interventions are worthy of further investigation in this context. In particular, there is promising evidence that ACT may improve the parenting of children with long-term conditions, seizure-control in epilepsy, psychological flexibility and possibly disease self-management/lifestyle.

A GENERAL CONCLUSION TO THE THESIS

Chapter 1 demonstrated that psychological flexibility is more strongly predictive of change in life satisfaction and anxiety over time than illness perceptions (with neither predictive of depression). Ostensibly, these findings suggest that interventions which focus on improving psychological flexibility may prove beneficial in muscle disorder care. However, the main caveat to this finding was that causality could not be established due mostly to the observational design. Therefore, it cannot be said that if one improves psychological flexibility then life satisfaction and anxiety will change as a result. Experimental methods which manipulate psychological flexibility are required to strengthen a case for causality.

ACT is a psychological therapy which is specifically designed to improve psychological flexibility. Given the findings of Chapter 1, we sought to examine its application in long-term conditions (comparable contexts to muscle disorders). This demonstrated that ACT has been applied in many ways to improve outcomes across conditions. However, despite encouraging results for some specific applications, the general quality of evidence was quite low and there were few RCT evaluations per application. Indeed, while improvements following ACT were seen for QoL and distress, there was little clear evidence that this was not due to regression to the mean or non-specific therapy factors. The implication for interventions with muscle disorders is that one cannot generalise that: ACT should improve these outcomes in muscle

disorders because this is clearly established in a similar context (i.e. with other long-term conditions). However, encouragingly the interventions did appear feasible and generally beneficial. Indeed, importantly the systematic review observed promising evidence that psychological flexibility may be changed as a result of ACT.

Therefore the findings and limitations of the two studies lead to one conclusion. In the context of muscle disorders which are associated with significant reductions in QoL (Burns et al., 2012; Graham et al., 2011) and for which medical treatment is limited, there is a suggestion that psychological flexibility might help explain variation in QoL/life satisfaction and mood. An RCT (of ACT) is an experimental design which will allow us to establish if psychological flexibility can be manipulated to improve outcomes, but more importantly if psychological intervention can improve outcomes in muscle disorders. With a lack of available generalisabile findings regarding efficacy from other long-term conditions, I would thus suggest that steps are now made towards undertaking such a trial with muscle disorders.

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Appendices

APPENDIX FOR CHAPTER ONE

(i) Exploratory Factor Analysis of Brief IPQ

Exploratory Factor Analysis of the Brief IPQ

Due to poor internal consistency the 8-item scale was subjected to a principal component analysis (Varimax rotation). The Kaiser-Meyer-Olkin measure (KMO = .63) suggested moderate sampling adequacy (Field, 2009), and KMO values for the individual items ranged from .49 - .70. Bartlett's test of Sphericity X^2 (137) = 167.98, p <.001, indicated that inter-correlations between items were acceptable for principal component analysis. Using Kaiser's criterion, three variables in combination explained 61.58% of the variance, points of inflection on the scree plot suggested retention of between one and three variables. Given that Kaiser's criterion lay within this range, three variables were extracted. These variables had internal consistency of α = .71 (emotional representation, consequences, concern and identity); α = .38 (coherence and personal control); and, α =.24 (timeline and treatment control). Factor 1 showed acceptable internal consistency, items were conceptually similar, and the concept of this item was commensurate with the planned overall scale (i.e. it appeared to measure illness threat as would be measured by the whole Brief IPQ scale). Thus factor 1 was retained in the analyses. Given that factors 2 and 3 lacked conceptual consistency and had poor internal consistency these were omitted.

Figure A1. Scree plot for the Brief IPQ

(ii) Assumptions of normality for regression analyses

Assumptions for regressions involving the SWLS

The assumptions for multiple regression as described by Field (2009) were considered. A brief description of each assumption, how it should be evaluated using SPSS, the performance of the present data and solutions are listed below.

Normally distributed dependent variables

Dependent variables must have at least an approximately normal distribution for regression analysis to be valid. Normality involves the distribution of responses across the possible data points of a given measure. Normal data follows a bell-curve distribution with greater frequency of response at central points. Skewness and kurtosis indicate non-normal distributions: kurtosis refers to the peakedness of the distribution,

whilst skewness regards the symmetry of the distribution. As suggested by Field (2009) we used histograms and normality plots to assess the normality of data. The Kolmogorov-Smirnov statistic was also inspected; however this may be over-sensitive to any violations of normality (Field, 2009), and therefore visual inspection of normality plots was the predominant measure. As you can see from the below histograms and normality plots (Table A1; Figures A2-A5), both the SWLS measured at time one, and the SWLS measured at time two had approximately normal distributions.

Table A1

Tests of Normality

	Kolmogorov-Smirnov[a]			Shapiro-Wilk		
	Statistic	df	Sig.	Statistic	df	Sig.
SWLS11	.093	137	.005	.970	137	.004
SWLST22	.077	137	.045	.966	137	.002

a. Lilliefors Significance Correction

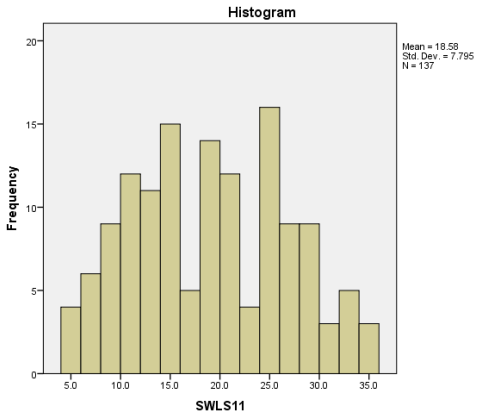


Figure A2

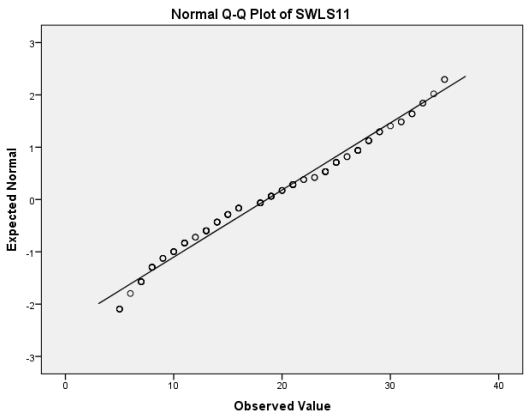


Figure A3

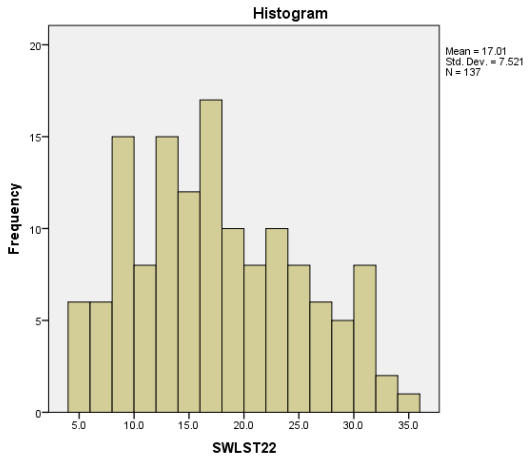


Figure A4

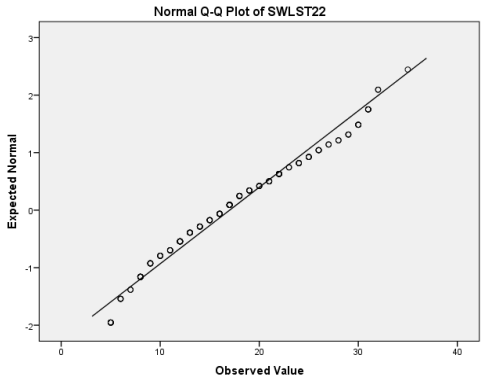


Figure A5

Normally distributed independent variables

Field (2009) suggests that due to central limit theorem, non-normally distributed independent variables are unlikely to invalidate the model in larger samples, but may influence smaller sample sizes. The present sample was of adequate sample size for the number of predictors; however, it could not be described as a large sample size (*N* =127). Thus non-normal predictor variables were transformed where possible. See below for normality indicators (Kolmogorov-Smirnov Test; histograms and normality plots of predictors), (Table A2; Figures A6-A15) and subsequent transformations (Table A3; Figures A16- A19).

Table A2

Tests of Normality

	Kolmogorov-Smirnov[a]			Shapiro-Wilk		
	Statistic	df	Sig.	Statistic	df	Sig.
HAQ-ALT	.105	137	.001	.930	137	.000
IPQ_CCIE11	.074	137	.064	.979	137	.031
AAQ9	.057	137	.200*	.992	137	.615
CFQ	.106	137	.001	.946	137	.000
ELSrecT	.070	137	.198	.968	137	.003

*. This is a lower bound of the true significance.

a. Lilliefors Significance Correction

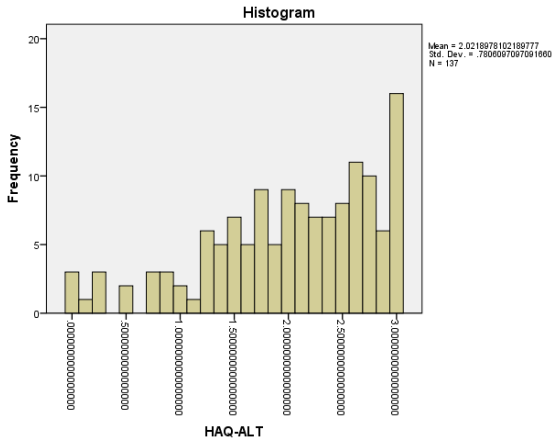


Figure A6

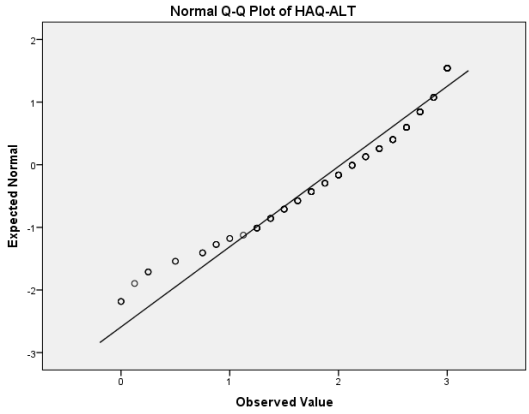


Figure A7

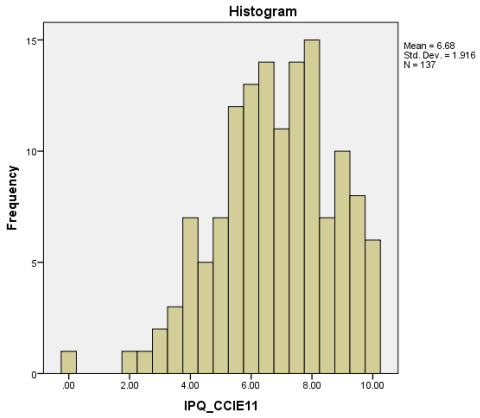


Figure A8

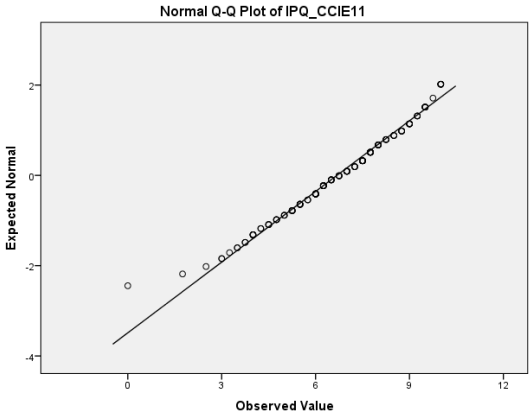


Figure A9

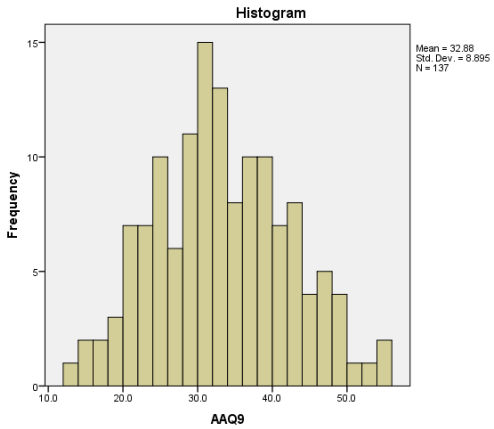


Figure A10

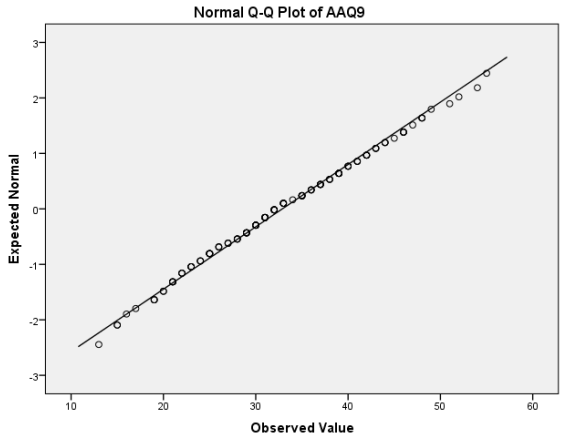


Figure A11

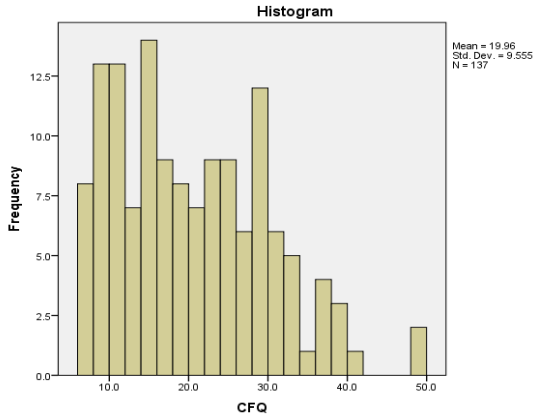


Figure A12

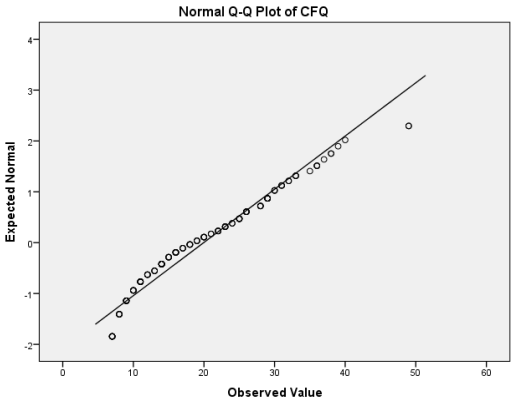


Figure A13

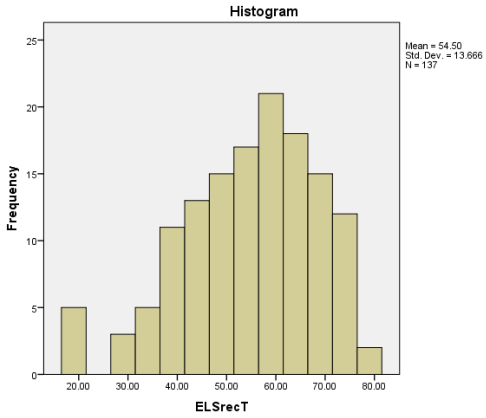


Figure A14

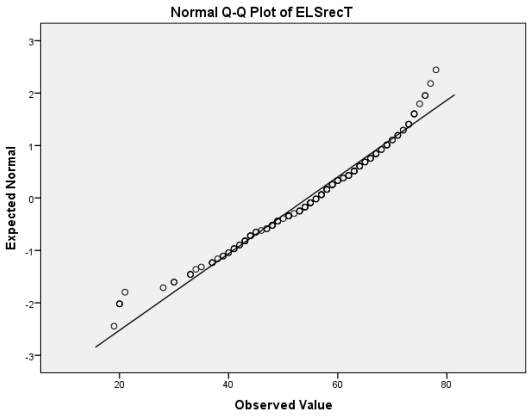


Figure A15

Given the non-normality displayed by the HAQ (negative skew) and CFQ (positive skew), several transformations were tried. A Log10 transformation gave the most normal distribution for the HAQ, while a square route transformation gave the most normal distribution for the CFQ; however, the distributions remained non-normal (Figures A16- A19).

Table A3

Tests of Normality

	Kolmogorov-Smirnov[a]			Shapiro-Wilk		
	Statistic	df	Sig.	Statistic	df	Sig.
HAQrLog10RRR	.085	137	.018	.962	137	.001
CFQ_Sqrt2	.084	137	.019	.967	137	.002

a. Lilliefors Significance Correction

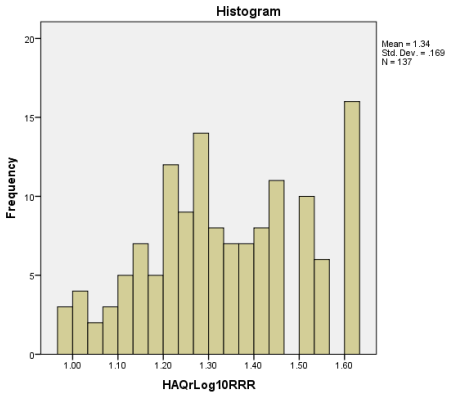


Figure A16

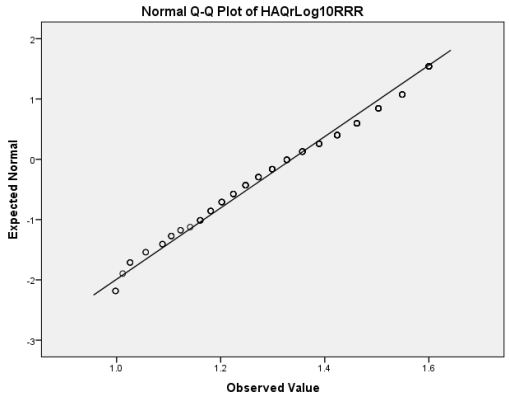


Figure A17

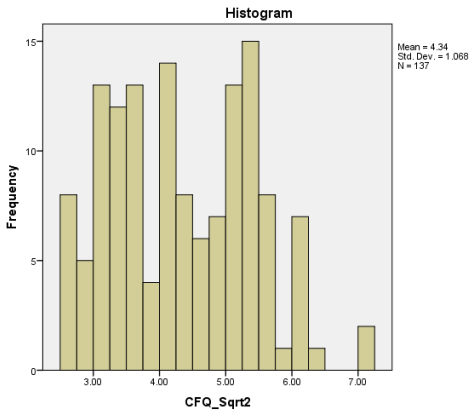


Figure A18

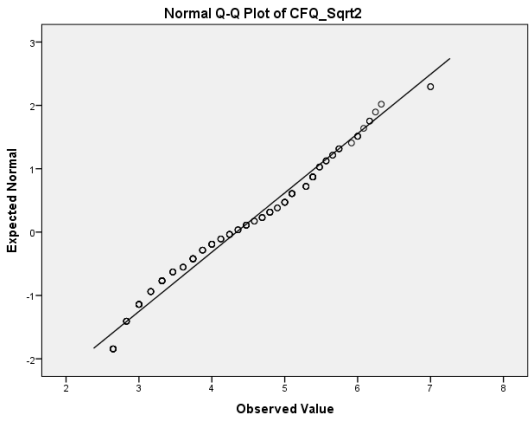


Figure A19

Linearity & Homoscedasticity

Multiple regression is based on the linear model. The assumption of linearity relates to whether the effects we are modelling can be described using this model (Field, 2009). It is also important that variance in residual terms should be should be equal at each level of the predictor variables (Field, 2009). This is called Homoscedasticity.

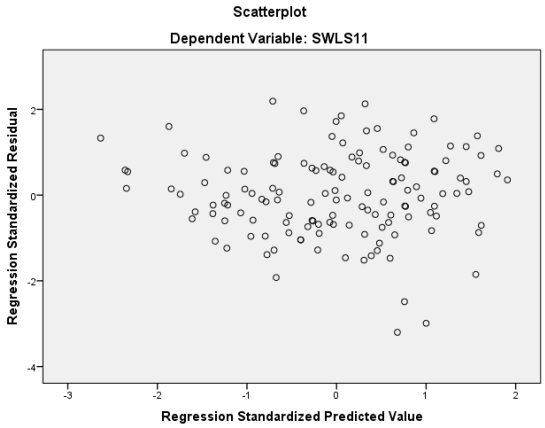


Figure A20

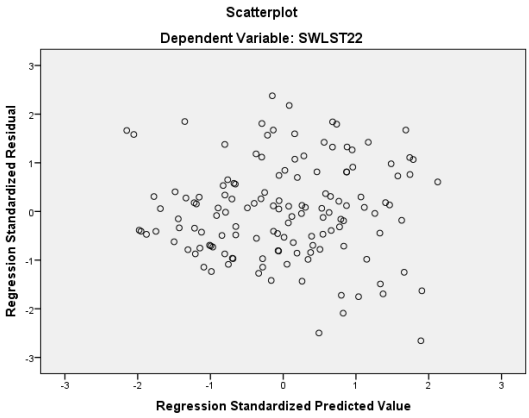


Figure A21

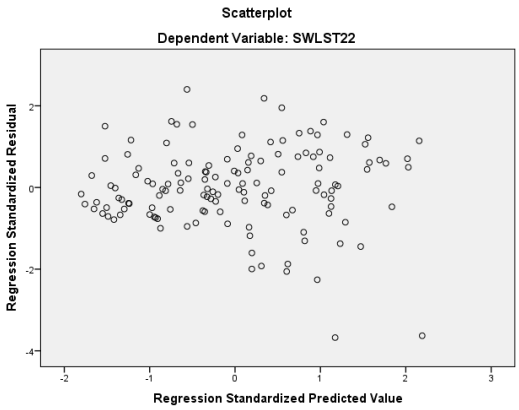


Figure A22

The plot for each of the three regressions is presented in Figures A20 – A22. Here, scatter plots revealed that assumptions of homoscedasticity (no funnelling of points on the graph) and linearity (no obvious curve pattern) were met for regressions 2 and 3 (Figure A21- A22)(Field, 2009). However there was some suggestion of homoscedasticity (slight funnelling of points on the graph) for regression 1. As this was not severe, it was expected that this should not bias the regression (Figure A20).

Independent errors

An association between the residual terms of any two observations also invalidates our model. This assumption was tested by using the Durbin-Watson test. This test statistic can range from 0-4. Field (2009) suggests that a value between one

and three is unlikely to be cause for concern. In regression one (with SWLS at time one as the dependent variable) the Durbin-Watson statistic was 1.87.

In regression two (with SWLS measured at time two as the dependent variable) the Durbin-Watson statistic was 2.00. In regression three (with SWLS measured at time two as the dependent variable and SWLS measured at time one included in the regression) the Durbin-Watson statistic was 2.42.

Normally distributed errors

Regression assumes that the residual terms are normally distributed, resulting in a mean of 0. As recommended by Field (2009), histograms and normality plots were inspected to assess whether this assumption was met. As illustrated in Fig A23-A28, the residual terms were approximately normally distributed.

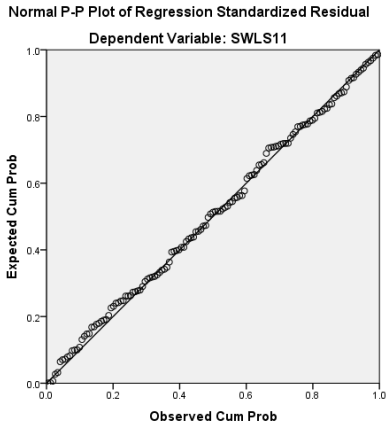


Figure A23

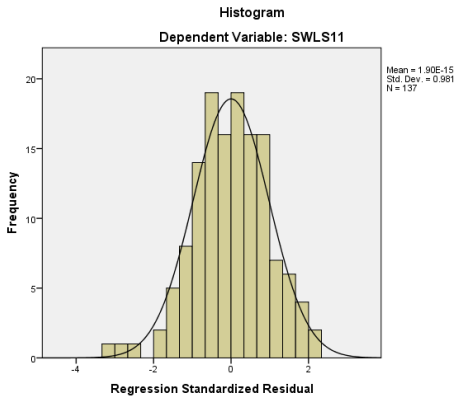
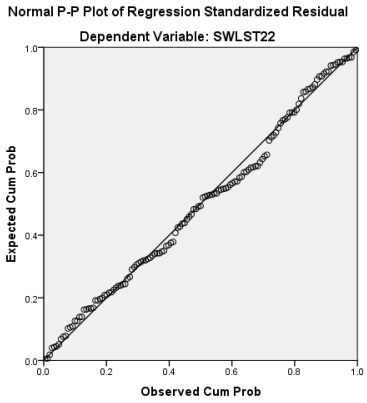


Figure A24



Fugure A25

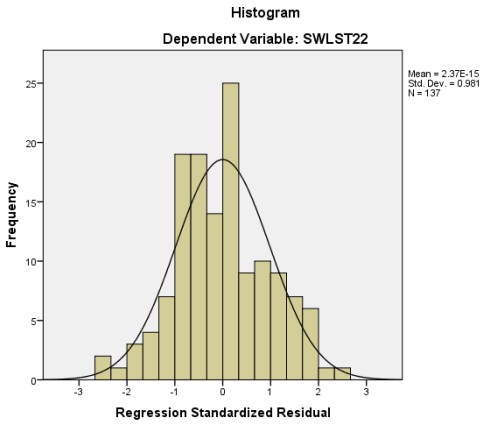


Figure A26

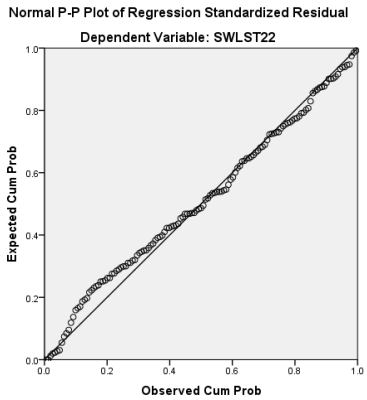


Figure A27

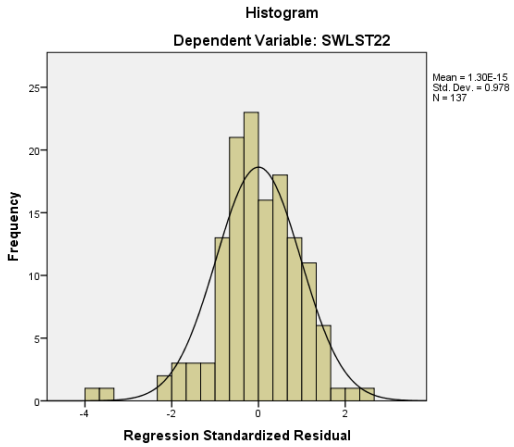


Figure A28

Multi-collinearity

Given that multiple regression was used, it is important that no perfect linear relationships are observed between the predictor variables. The variance inflation factor (VIF), which assesses the strength of the relationship between each individual predictor and the other predictors, was used to assess multi-collinearity. The largest VIF should be less than 10 and the average VIF should not be substantially greater than 1 (Field, 2009). A tolerance statistic, dervived from the VIF, should also remain above 0.2 (Field, 2009).

In regression one (with SWLS measured at time one as the dependent variable) VIF ranged from 1.08– 2.23, average VIF was 1.33, with tolerance ranging from 0.45 – 0.93. Regression two (with SWLS measured at time two as the dependent variable) VIF ranged from 1.00 – 2.23, average VIF was 1.73, with tolerance ranging from 0.45 – 0.93. In regression three (with SWLS measured at time two as the dependent variable and SWLS measured at time one included in the regression) VIF ranged from 1.00 – 3.71, average VIF was 2.11, with tolerance ranging from 0.33 -0.91

Outliers

Field (2009) suggests that regression analysis is sensitive to outliers. In the present analysis we used Mahalanobis distance (the distance of a case from the means of

all other cases, with the centriod created using the means of all standardised variables [Field, 2009]) to assess for the presence of outliers. Using a a table of critical values derived from number of predictors and sample size, (Bartnett & Lewis 78), it was established that distances of greater than 22 were likely to indicate an influential outlier in the present sample. In regression one the greatest Mahalanobis distance was 20.71. In regression two the greatest Mahalanobis distance was 20.71. In regression three, the greatest Mahalanobis distance was 20.91.

Assumptions for regressions involving the GAD-7

This appendix details the results of the preliminary analysis for regressions where the GAD-7 was the dependent variable.

Normally distributed dependent variables

As you can see from the below histograms and normality plots, both the GAD-7 measured at time one, and the GAD-7 measured at time two had a non- normal distribution, with a positive skew (Table A4; Figures A29-32). Several transformations were tried, but only an approximate transformation was possible (Square route) for both variables (Table A5; Figures A33-36). As recommended by Field (2009), given that a non-normal distribution can affect the validity of results, robust regression procedures were used in all regressions where GAD-7 was the dependent variable.

Table A4

Tests of Normality

	Kolmogorov-Smirnov[a]			Shapiro-Wilk		
	Statistic	df	Sig.	Statistic	df	Sig.
GAD_TOT11	.165	137	.000	.834	137	.000
GADTot22	.176	137	.000	.853	137	.000

a. Lilliefors Significance Correction

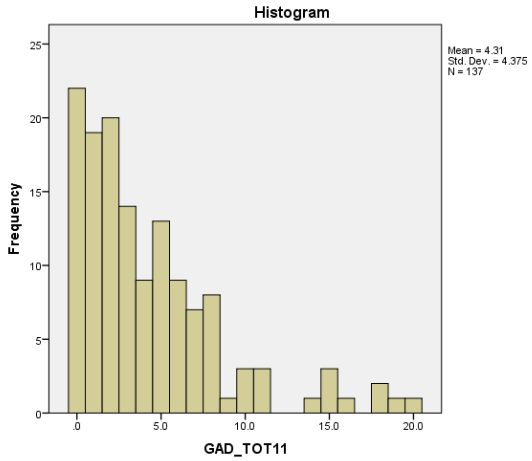


Figure A29

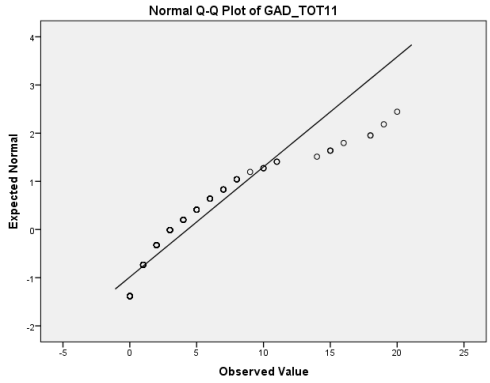


Figure A30

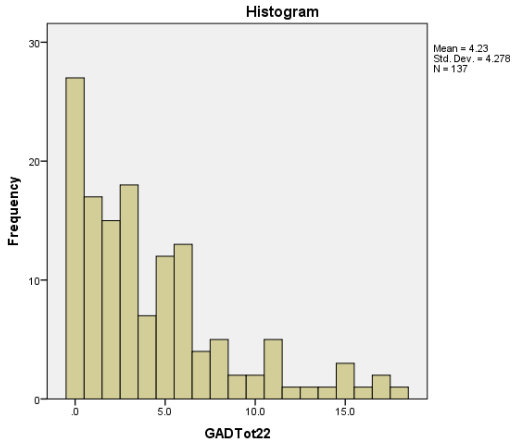


Figure A31

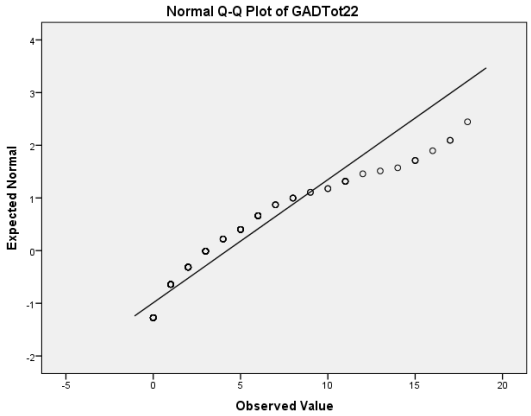


Figure A32

Table A5

Tests of Normality

	Kolmogorov-Smirnov[a]			Shapiro-Wilk		
	Statistic	df	Sig.	Statistic	df	Sig.
GAD_SQ	.104	137	.001	.956	137	.000
GAD2_SQ	.130	137	.000	.946	137	.000

a. Lilliefors Significance Correction

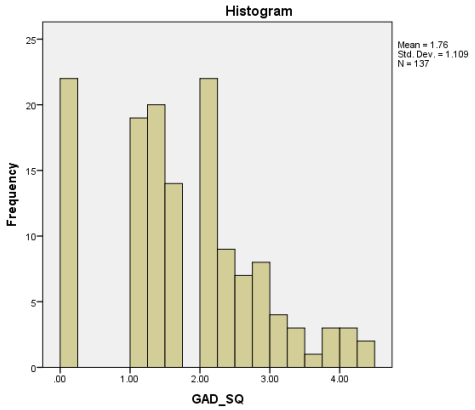


Figure A33

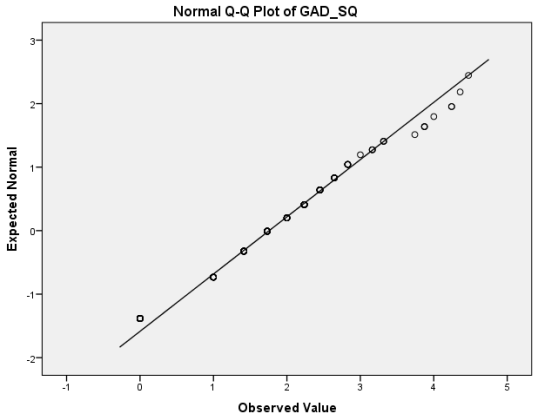


Figure A34

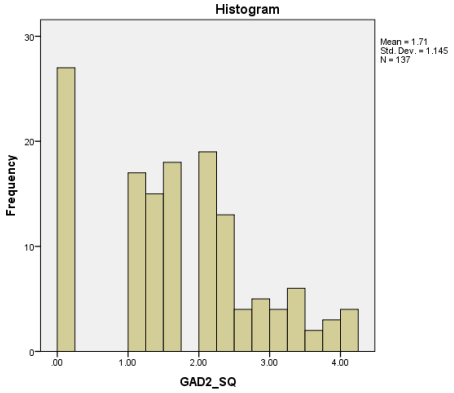


Figure A35

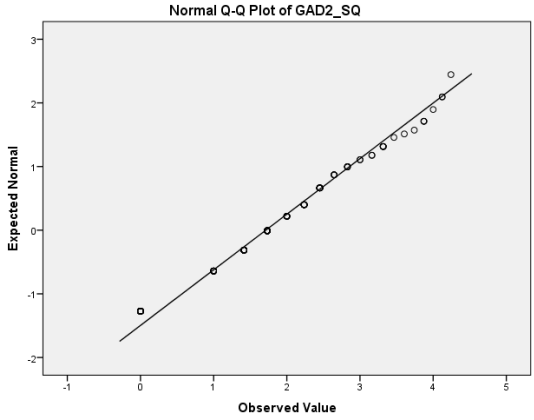


Figure A36

Normally distributed independent variables

The previous section (assumptions for SWLS regressions) outlines the normality of predictor variables and subsequent transformations.

Linearity & Homoscedasticity

The scatter plots (Figures A37-A39) revealed that assumptions of homoscedasticity (no funnelling of points on the graph) and linearity (no obvious curve pattern) were met (Field, 2009).

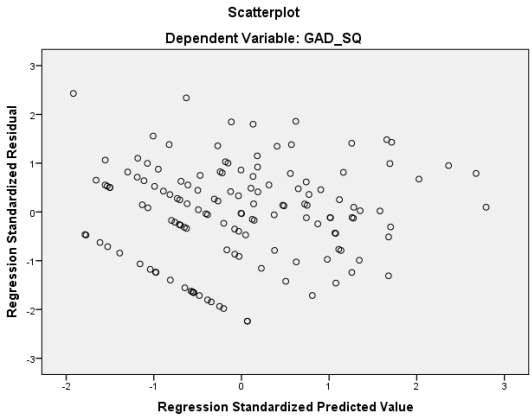


Figure A37

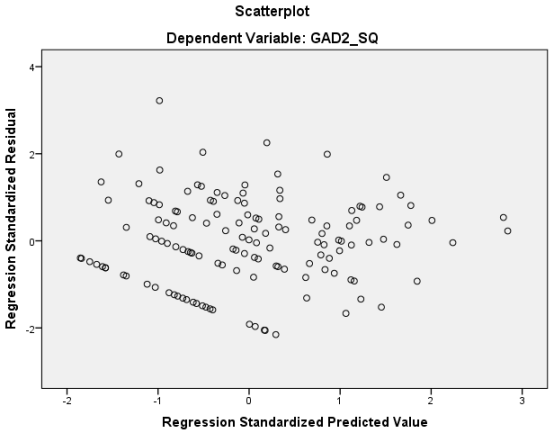


Figure A38

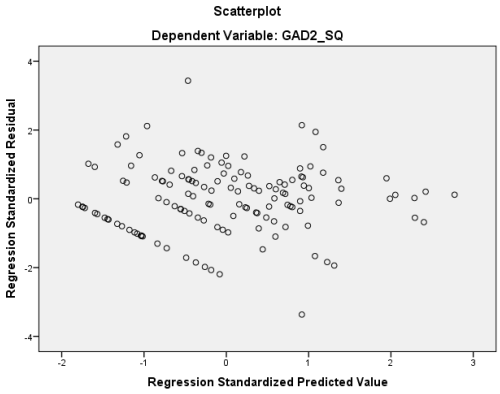


Figure A39

Independent errors

In regression one the Durbin-Watson statistic was 1.85. In regression two the Durbin-Watson statistic was 2.24. In regression three the Durbin-Watson statistic was 2.09.

Normally distributed errors

Regression assumes that the residual terms are normally distributed, resulting in a mean of 0. As recommended by Field (2009), histograms and normality plots were

inspected to assess whether this assumption was met. As illustrated in Figures A40-A45 these were approximately normally distributed.

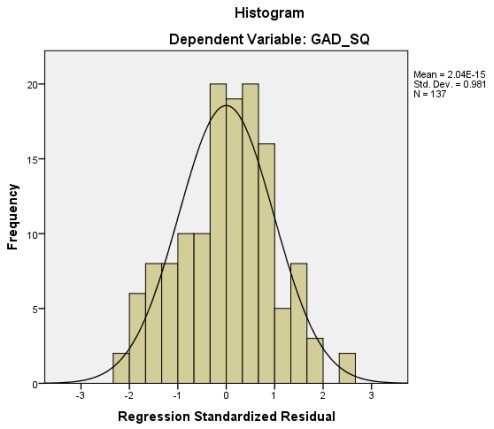


Figure A40

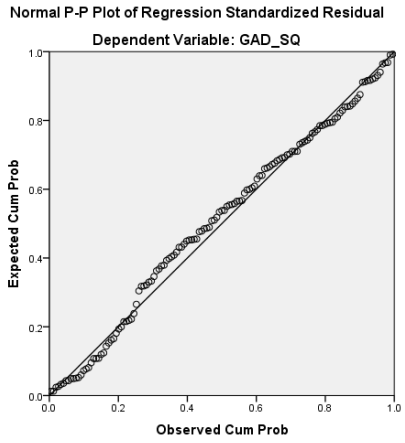


Figure A41

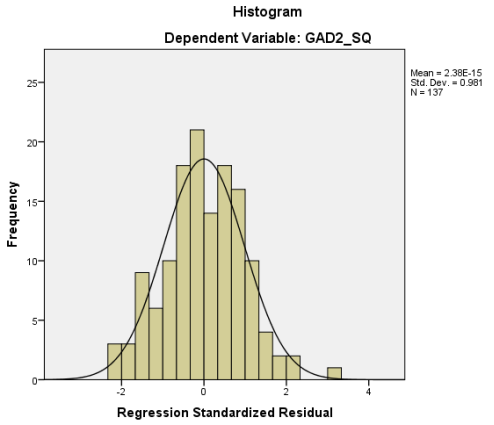


Figure A42

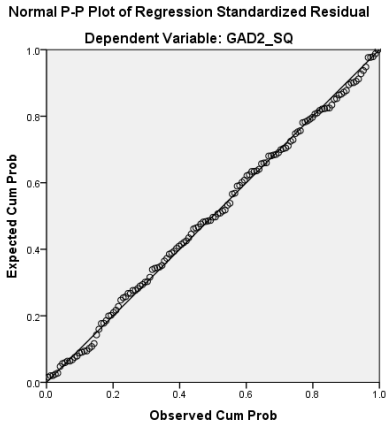


Figure A43

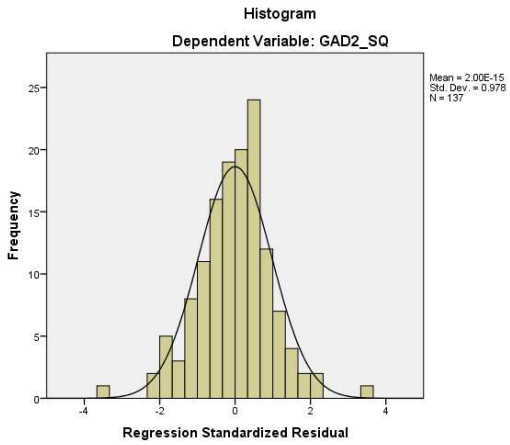


Figure A44

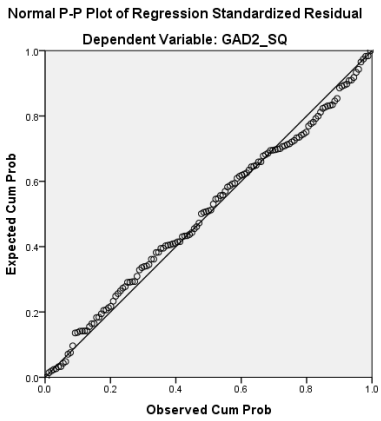


Figure A45

Multi-collinearity

In regression one VIF ranged from 1.27– 1.41, average VIF was 1.33, with tolerance ranging from 0.71 – 0.79. In regression twoVIF ranged from 1.08 – 2.23, average VIF was 1.73, with tolerance ranging from 0.45 – 0.93. In regression three VIF ranged from 1.08 – 2.24, average VIF was 1.85, with tolerance ranging from 0.45 - 0.92.

Outliers

In regression one the greatest Mahalanobis distance was 20.71. In regression two the greatest Mahalanobis distance was 20.71. In regression three the greatest Mahalanobis distance was 21.34.

Assumptions for regressions involving the PHQ-9

This appendix details the results of the preliminary analysis for regressions where the PHQ-9 was used as the dependent variable. The assumptions for multiple regression as described by Field (2009) are listed below.

Normally distributed dependent variables

As illustrated in the below histograms and normality plots, both the PHQ-9 measured at time one, and the PHQ-9 measured at time two had non- normal distributions, with a positive skew (Table A6; Figures A46-49). Several transformations were tried, but only an approximate transformation was possible (Square route) for both variables (Table A7 Figures A50-53). As recommended by Field (2009), given that a

non-normal distribution can affect the validity of results, robust regression procedures were used in all regressions where PHQ-9 was the dependent variable.

Table A6

Tests of Normality

	Kolmogorov-Smirnov[a]			Shapiro-Wilk		
	Statistic	df	Sig.	Statistic	df	Sig.
PHQTOT11	.137	137	.000	.920	137	.000
PHQTot	.131	137	.000	.925	137	.000

a. Lilliefors Significance Correction

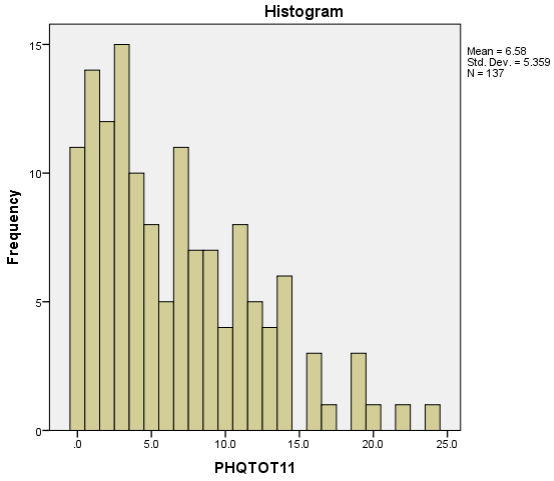


Figure A46

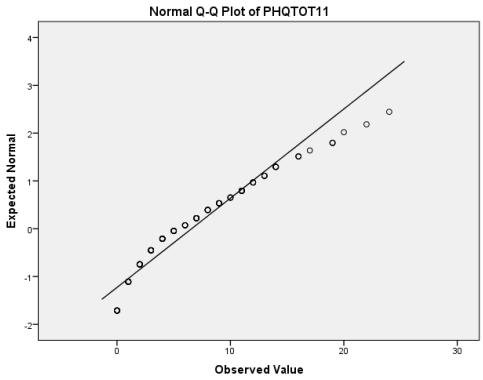


Figure A47

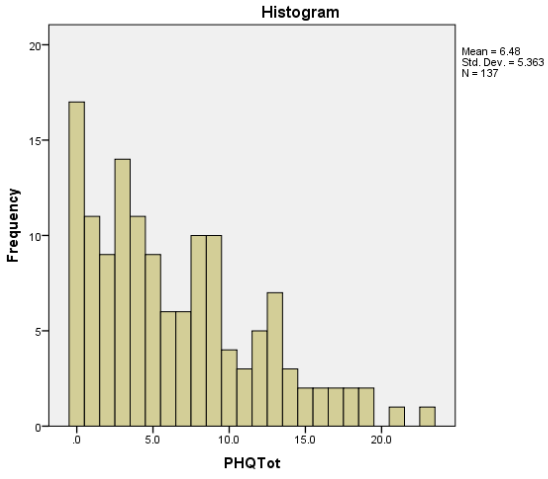


Figure A48

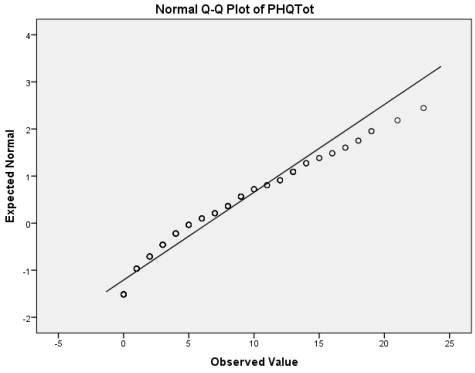


Figure A49

Table A7

Tests of Normality

	Kolmogorov-Smirnov[a]			Shapiro-Wilk		
	Statistic	df	Sig.	Statistic	df	Sig.
PHQ_SQ	.073	137	.069	.975	137	.014
PHQ2_SQ	.090	137	.009	.961	137	.001

a. Lilliefors Significance Correction

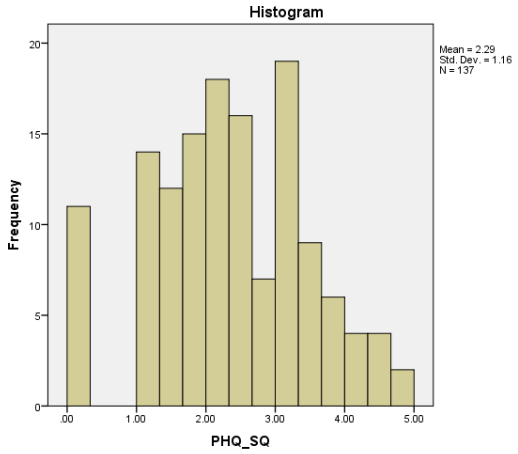


Figure A50

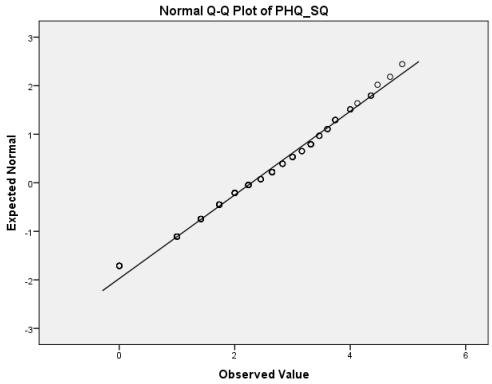


Figure A51

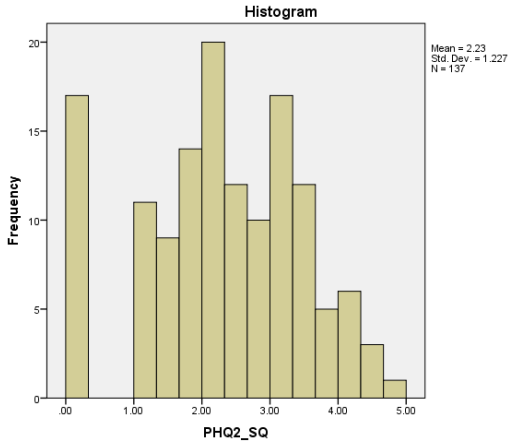


Figure A52

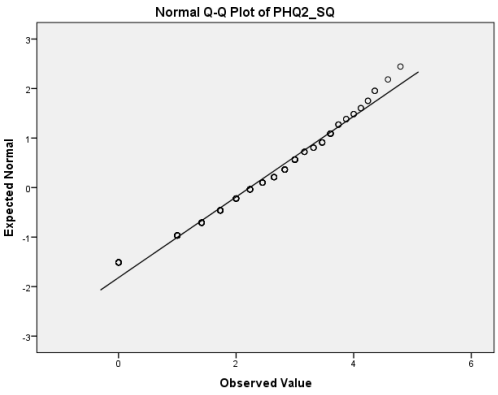


Figure A53

Normally distributed independent variables

See the previous section (assumptions for regressions involving the SWLS) which outlines the normality of independent variables and subsequent transformations.

Linearity & Homoscedasticity

The scatter plots revealed that assumptions of homoscedasticity (no funnelling of points on the graph) and linearity (no obvious curve pattern) were met (Field, 2009) (Figures A54-A56).

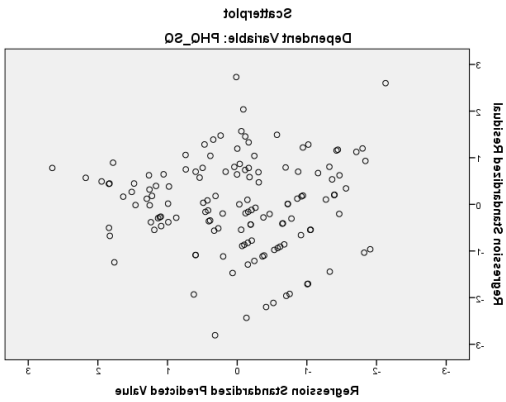


Figure A54

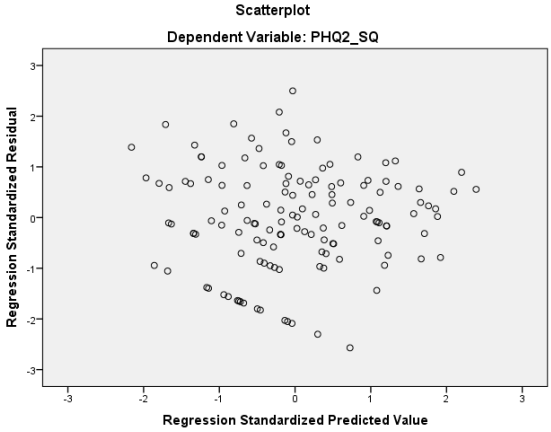


Figure A55

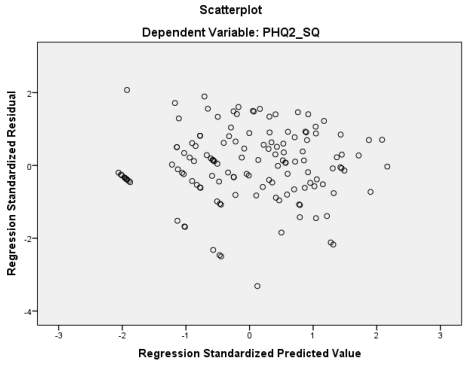


Figure A56

Independent errors

In regression one the Durbin-Watson statistic was 2.11. In regression two the Durbin-Watson statistic was 2.23. In regression three the Durbin-Watson statistic was 2.17.

Normally distributed errors

Histograms and normality plots were inspected to assess whether this assumption was met. As illustrated in Figures A57-A62 these were all approximately normal.

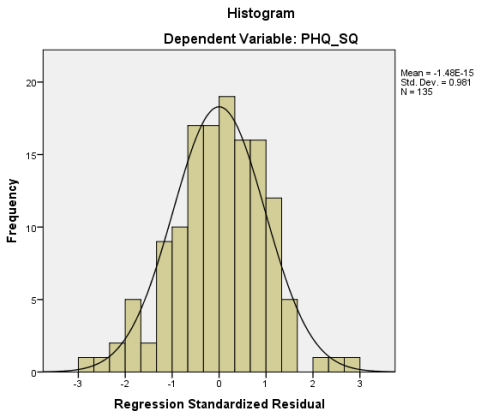


Figure A57

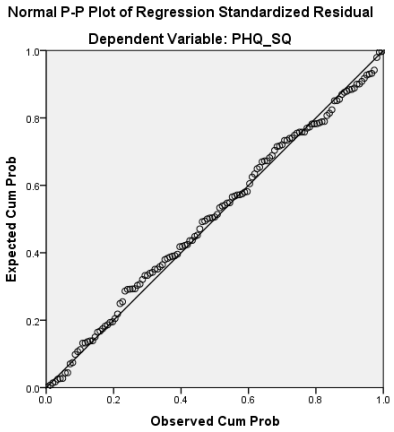


Figure A58

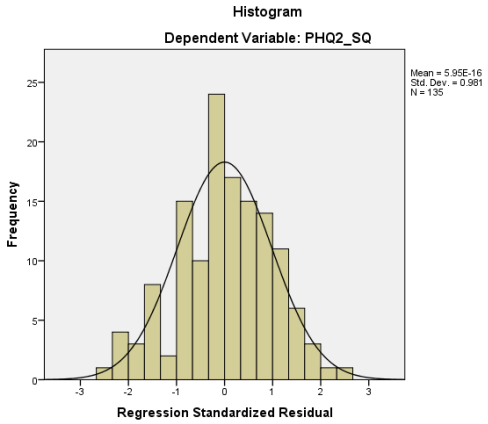


Figure A59

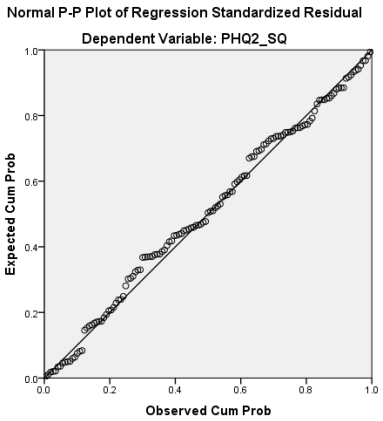


Figure A60

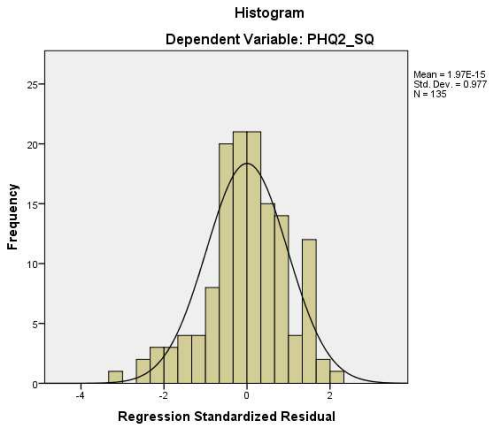


Figure A61

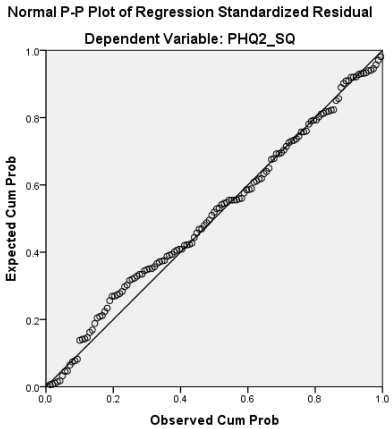


Figure A62

Multi-collinearity

In regression one VIF ranged from 1.08– 2.07, average VIF was 1.63, with tolerance ranging from 0.48 – 0.92. In regression two VIF ranged from 1.08 – 2.07, average VIF was 1.63, with tolerance ranging from 0.48 – 0.92. In regression three VIF ranged from 1.08 – 2.08, average VIF was 1.70, with tolerance ranging from 0.52 -0.92.

Outliers

In regression one the greatest Mahalanobis distance was 21.02. In regression two the greatest Mahalanobis distance was 21.02. In regression three the greatest Mahalanobis distance was 22.07.

Additional References

Barnett, V., & Lewis, T. (1978). Outliers in statistical data. New York: Wiley

Field A. (2013). *Discovering statistics using IBM SPSS statistics.* London: Sage Publications.

(iii) Extended versions of the regression tables

Table a. Three regressions showing the cross-sectional and prospective influence of independent variables on life satisfaction (SWLS).

A. Crossectional: all variables at T1

Step	Variable	β	t	p	R^2	ΔR^2	ΔF	p
1	Disability Level (HAQ-DI)	-.11	-1.29	.20	.01	.01	1.68	.20
2	Disability Level (HAQ-DI)	.01	0.11	.92	.27	.26	47.92	<.001
	Illness Threat (IPQ-Threat)	-.52	-6.92	<.001				
3	Disability Level (HAQ-DI)	-.08	-1.47	.14	.62	.34	39.22	<.001
	Illness Threat (IPQ-Threat)	-.22	-3.27	.001				
	Experiential Avoidance (AAQ)	-.10	-1.22	.23				
	Cognitive Fusion (CFQ)	.13	1.81	.07				
	Valued-living (ELS)	.66	8.75	<.001				

B. Prospective: Independent variables at T1; Dependent variables at T2

Step	Variable	β	t	p	R^2	ΔR^2	ΔF	p
1	Disability Level (HAQ-DI)	-.10	-1.19	.27	.01	.01	1.25	.27
2	Disability Level (HAQ-DI)	.01	0.09	.94	.20	.19	32.34	<.001
	Illness Threat (IPQ-Threat)	-.45	-5.69	<.001				
3	Disability Level (HAQ-DI)	-.07	-0.98	.33	.43	.23	17.65	<.001
	Illness Threat (IPQ-Threat)	-.23	-2.79	.01				
	Experiential Avoidance (AAQ)	-.28	-2.84	.01				
	Cognitive Fusion (CFQ)	.30	3.34	.01				
	Valued-living (ELS)	.45	4.89	<.001				

C. Prospective: Independent variables at T1; Dependent variables at T2 controlling for T1.

Step	Variable	β	t	p	R^2	ΔR^2	ΔF	p
1	T1 Life Satisfaction (SWLS)	.72	12.14	<.001	.52	.52	147.31	<.001
2	T1 Life Satisfaction (SWLS)	.72	11.99	<.001	.52	.00	0.72	.789
	Disability Level (HAQ-DI)	-.02	-.27	.79				
3	T1 Life Satisfaction (SWLS	.67	9.61	<.001	.52	.01	1.95	.165
	Disability Level (HAQ-DI)	.00	0.02	.99				
	Illness Threat (IPQ-Threat)	-.10	-1.40	.17				

4	T1 Life Satisfaction (SWLS)	.59	6.33	<.001	.55	.04	3.65	.014
	Disability Level (HAQ-DI)	-.02	-0.30	.77				
	Illness Threat (IPQ-Threat)	-.10	-1.32	.19				
	Experiential Avoidance (AAQ)	-.22	-2.55	.01				
	Cognitive Fusion (CFQ)	.22	2.78	.01				
	Valued-living (ELS)	.06	0.58	.57				

Method: Enter

SWLS= Satisfaction with Life Scale; HAQ-DI = Health Assessment Questionnaire – Disability Index; IPQ Threat = Brief Illness Perception Questionnaire Threat Scale; AAQ = Acceptance and Action Questionnaire; CFQ = Cognitive Fusion Questionnaire; ELS = Engaged Living Scale.

Table b. Three regressions showing the cross-sectional and prospective influence of independent variables on anxiety (GAD 7)†

A. Crossectional: all variables at T1

Step	Variable	β	t	p	R^2	ΔR^2	ΔF	p
1	Disability Level (HAQ-DI)	.02	0.25	.80	-.01	.00	0.06	.801
2	Disability Level (HAQ-DI)	-.10	-1.26	.25	.25	.26	46.18	<.001
	Illness Threat (IPQ-Threat)	.52	6.80	<.001				
3	Disability Level (HAQ-DI)	-.04	-0.59	.60	.47	.23	19.75	<.001
	Illness Threat (IPQ-Threat)	.20	-2.58	.02				
	Experiential Avoidance (AAQ)	.03	-0.35	.69				
	Cognitive Fusion (CFQ)	.38	4.50	<.001				
	Valued-living (ELS)	-.23	-2.67	.016				

B. Prospective: Independent variables at T1; Dependent variables at T2

Step	Variable	β	t	p	R^2	ΔR^2	ΔF	p
1	Disability Level (HAQ-DI)	-.07	-0.80	.42	.01	.01	0.64	.43
2	Disability Level (HAQ-DI)	-.17	-2.10	.03	.19	.18	30.09	<.001
	Illness Threat (IPQ-Threat)	.44	5.49	.00				
3	Disability Level (HAQ-DI)	-.11	-1.52	.15	.41	.22	16.53	<.001
	Illness Threat (IPQ-Threat)	.12	1.42	.27				
	Experiential Avoidance (AAQ)	-.06	0.64	.56				
	Cognitive Fusion (CFQ)	.31	3.42	.01				
	Valued-living (ELS)	-.27	-2.92	<.01				

C. Prospective: Independent variables at T1; Dependent variables at T2 controlling for T1.

Step	Variable	β	t	p	R^2	ΔR^2	ΔF	p
1	T1 Anxiety (GAD-7)	.75	12.96	<.001	.56	.56	168.07	<.001
2	T1 Anxiety (GAD-7)	.75	13.05	<.001	.56	.01	2.20	.14
	Disability Level (HAQ-DI)	-.09	-1.48	.17				
3	T1 Anxiety (GAD-7)	.71	10.75	<.001	.57	.00	1.00	.32
	Disability Level (HAQ-DI)	-.10	-1.69	.11				
	Illness Threat (IPQ-Threat)	.07	1.00	.31				
4	T1 Anxiety (GAD-7)	.59	7.58	<.001	.59	.03	2.78	.04
	Disability Level (HAQ-DI)	-.08	-1.42	.17				

Illness Threat (IPQ-Threat)	.00	-0.01	.99
Experiential Avoidance (AAQ)	.05	-0.53	.67
Cognitive Fusion (CFQ)	.08	1.03	.35
Valued-living (ELS)	-.14	-1.68	.11

Method: Enter

GAD-7= General Health Questionnaire 7 item version; HAQ-DI = Health Assessment Questionnaire – Disability Index; IPQ Threat = Brief Illness Perception Questionnaire Threat Scale; AAQ = Acceptance and Action Questionnaire; CFQ = Cognitive Fusion Questionnaire; ELS = Engaged Living Scale.

† Bootstrapped p-values reported

Table c. Three regressions showing the cross-sectional and prospective influence of independent variables on depression (PHQ 9) †.

A. Crossectional: all variables at T1

Step	Variable	β	t	p	R^2	ΔR^2	ΔF	p
1	Disability Level (HAQ-DI)	.08	0.97	.33	.01	.01	0.95	.332
2	Disability Level (HAQ-DI)	-.04	-0.50	.61	.28	.27	50.53	<.001
	Illness Threat (IPQ-Threat)	.54	7.11	<.001				
3	Disability Level (HAQ-DI)	.02	0.31	.77	.45	.17	13.06	<.001
	Illness Threat (IPQ-Threat)	.26	3.16	.02				
	Experiential Avoidance (AAQ)	.08	0.87	.34				
	Cognitive Fusion (CFQ)	.20	2.22	.09				
	Valued-living (ELS)	-.28	-3.10	<.01				

B. Prospective: Independent variables at T1; Dependent variables at T2

Step	Variable	β	t	p	R^2	ΔR^2	ΔF	p
1	Disability Level (HAQ-DI)	.06	0.71	.49	.00	.00	0.50	.48
2	Disability Level (HAQ-DI)	-.04	-0.50	.62	.19	.19	30.79	<.001
	Illness Threat (IPQ-Threat)	.44	5.55	.00				
3	Disability Level (HAQ-DI)	.01	0.15	.87	.31	.12	7.79	<.001
	Illness Threat (IPQ-Threat)	.19	2.08	.06				
	Experiential Avoidance (AAQ)	.18	1.66	.06				
	Cognitive Fusion (CFQ)	.16	1.60	.18				
	Valued-living (ELS)	-.16	-1.59	.11				

C. Prospective: Independent variables at T1; Dependent variables at T2 controlling for T1.

Step	Variable	β	t	p	R^2	ΔR^2	ΔF	p
1	T1 Depression (PHQ-9)	.83	17.06	<.001	.68	.68	291.06	<.001
2	T1 Depression (PHQ-9)	.83	16.95	<.001	.68	.00	0.03	.86
	Disability Level (HAQ-DI)	-.01	-0.17	.88				
3	T1 Depression (PHQ-9)	.83	14.39	<.001	.68	.00	0.00	.99
	Disability Level (HAQ-DI)	-.01	-0.17	.87				
	Illness Threat (IPQ-Threat)	.00	0.00	.99				
4	T1 Depression (PHQ-9)	.82	12.56	<.001	.69	.01	0.84	.47
	Disability Level (HAQ-DI)	-.01	-0.11	.91				

Illness Threat (IPQ-Threat)	-.02	-0.36	.70
Experiential Avoidance (AAQ)	.11	-1.50	.12
Cognitive Fusion (CFQ)	-.01	-0.07	.96
Valued-living (ELS)	.07	1.01	.37

Method: Enter

PHQ-9= Patient Health Questionnaire 9-item version; HAQ-DI = Health Assessment Questionnaire – Disability Index; IPQ Threat = Brief Illness Perception Questionnaire Threat Scale; AAQ = Acceptance and Action Questionnaire; CFQ = Cognitive Fusion Questionnaire; ELS = Engaged Living Scale.

† Bootstrapped p-values reported

(iv) Online materials

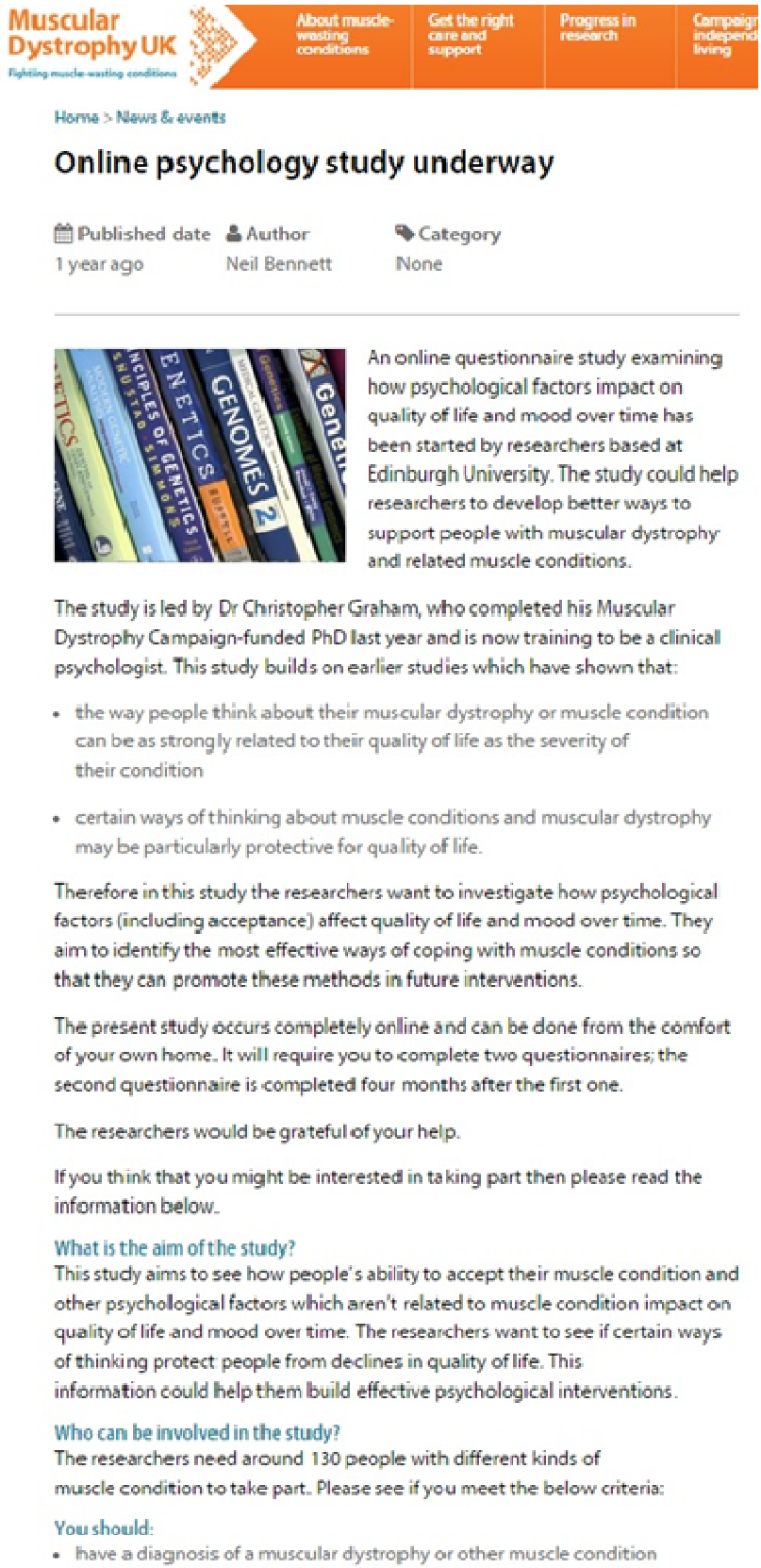


Figure B1. Online advertisement

A questionnaire study examining how psychological factors impact on quality of life and mood in people with muscle disease

Information Sheet

WELCOME and thanks for coming! Please read the below information about the study before you go any further...

INTRODUCTION

This study builds on earlier studies which have shown that: 1) the way people think about their muscular dystrophy can be as strongly related to their quality of life as disease severity; 2) certain ways of thinking (or behaving) about muscle disease based around acceptance may be particularly protective for quality of life.
Therefore in this study we want to investigate how psychological factors (including acceptance) affect quality of life and mood over time. The aim is to help us identify the most effective ways of coping with muscle disease, so that we can promote these methods in future interventions.

The present study occurs completely online and can be done from the comfort of your own home. It will require you to complete two questionnaires; the second questionnaire is completed four months after the first one.

If you think that you might be interested in taking part then please read the information below.

WHAT IS THE PURPOSE OF THIS STUDY?

This study aims to see how people's ability to accept their muscle disease and other psychological factors which aren't related to muscle disease impact on quality of life and mood over time. We want to see if certain ways of thinking protect people from declines in quality of life. This information can help us build effective psychological interventions.

If you are interested then please see if you fit the following criteria:

You should:
1. Have a diagnosis of a muscular dystrophy/ muscle disease.
2. Have a duration of muscular dystrophy/muscle disease of greater than six months.
3. Be aged between 18 and 75 years.
4. Be able to read in English.
5. Have access to the internet and a computer on which you can complete the questionnaires.

You should not:
1. Have major active illnesses which are unrelated to MD such as arthritis, respiratory disease, stroke.
2. Have a diagnosis of myotonic dystrophy.
3. Have cognitive impairment (i.e.significant memory/attention problems) e.g. from a stroke or Alzheimer's Disease.
4. Have a major diagnosed active mental health condition e.g. psychosis, major depression, obsessive compulsive disorder.
5. Be currently participating in any treatment intervention studies.

DO I HAVE TO TAKE PART IN THE STUDY?

Your participation is voluntary and even if you do start the study you may withdraw from the study at any time.

WHAT WILL HAPPEN TO ME IF I TAKE PART AND WHAT STUDY PROCEDURES AND TESTS WILL BE INVOLVED?

You will be asked to complete two questionnaires; both of these questionnaires will be completed online so you can do these from your own home.

Figure B2.Part of the Information sheet

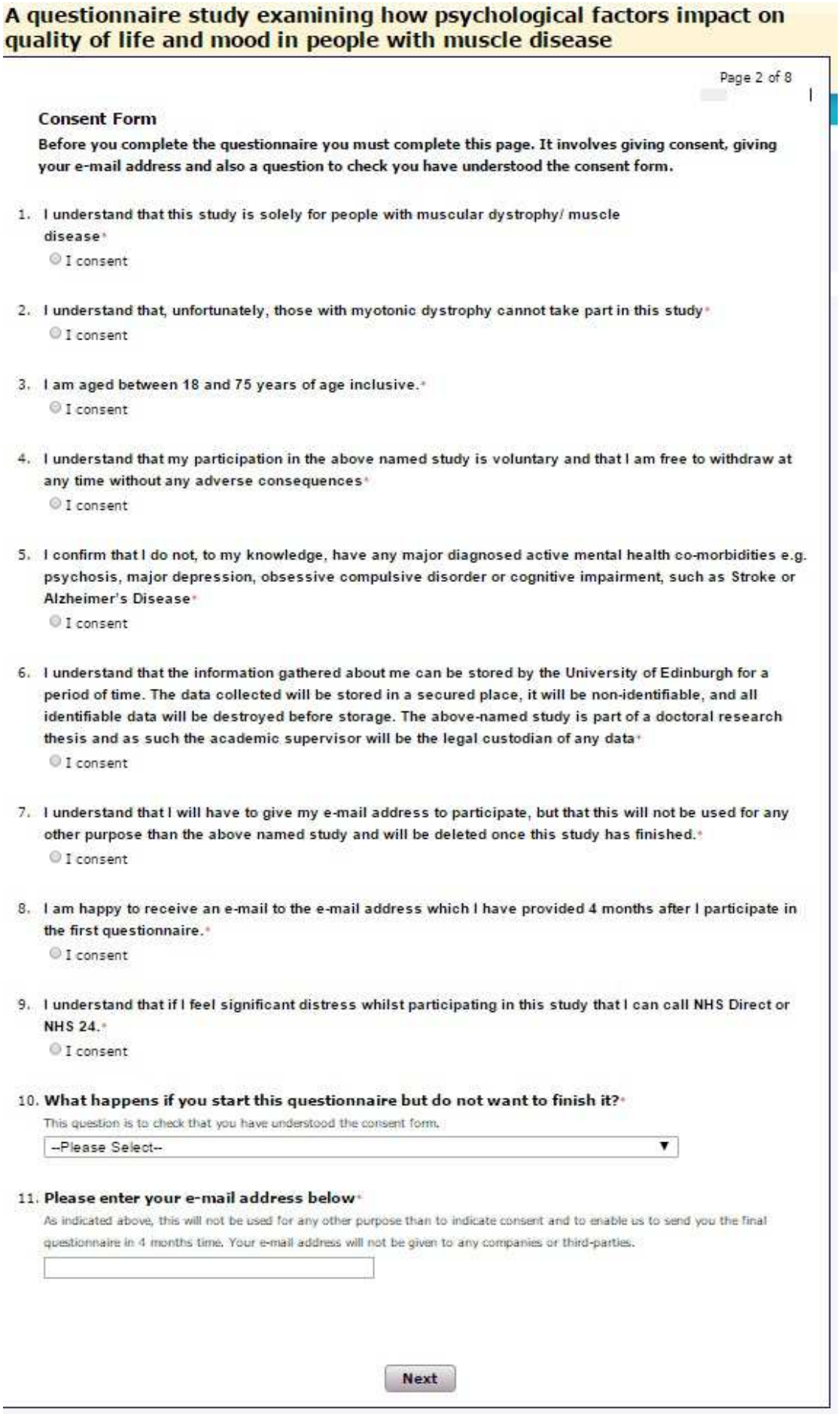


Figure B3. The consent form

(v) Redacted Ethics Approval Form

University of Edinburgh, School of Health in Social Science

RESEARCH ETHICS APPLICATION (REA)

The forms required when seeking ethical approval in the School of Health and Social Sciences have now been merged into this single electronic document. The sections you are required to complete will depend on the nature of your application. Please start to complete the form from the beginning and proceed as guided. On completion the ***entire*** document should be submitted electronically to your section's ethics tutor using the email addresses detailed on the final page.

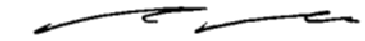
ER35 ISSUES ARISING FROM THE PROPOSAL

I can confirm that the above application has been reviewed by two independent reviewers. It is their opinion that:

a) The ethical issues listed below arise or require clarification:

- In the advert , the website for the questionnaire should only appear at the end. Currently there is a link provided half-way through the document, this should be removed.
- An independent advisor to the study contact could be provided both in the advert and on the participant information sheet.
- The consent form should make evident that data collected will be stored in a secured place, that it will be non-identifiable, and that all identifiable data will be destroyed.
- We ask if there is a more suitable source of help to direct participants in distress too that NHS 24 / Direct? These helplines have limited capacity to deal with psychological distress.

The applicant should respond to these comments in section 8 below.

Signature: 

Position: **Ethics Tutor**

Date: 14.11.2013

ER36 APPLICANT'S RESPONSE (If required)

Dear Reviewers,

Thank you for your detailed feedback on this ethics application, and apologies that it was an overly-long application. Below I list how I have addressed each of the issues that you specify above:

1. The link has been removed from the middle of the advert and placed at the end.
2. An independent point of contact has been placed on the information sheet and advert.
3. The consent form has been modified to include your suggestion.
4. We understand your concern regarding the use of the NHS 24 telephone number, however having considered this point, we think it is important that this contact number remains in the information sheets/thank you message. This is because, whilst we are aware that NHS 24 cannot directly treat any immediate distress, they will be able to suggest appropriate courses of action given the need of the caller - thus in a sense triage the level of need. Indeed this has been found acceptable by NHS ethics committees in my own previous NHS ethics applications. We will however provide a link to the mental health helpline which are suggested by the NHS (http://www.nhs.uk/Conditions/stress-anxiety-depression/Pages/mental-health-helplines.aspx) to allow participants to access other services under their own volition, if they so wish. I hope that this course of actions helps attenuate your concerns.

Thank you again for taking time to look through our application in detail,

Kind regards,

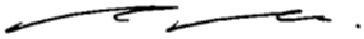
Chris Graham

Signature: 

Date:08.12.13

ER37 **CONCLUSION TO ETHICAL REVIEW (if required)**

The applicant's response to our request for further clarification or amendments has now satisfied the requirements for ethical practice and the application has therefore been approved.

signature: 

Position: Ethics Tutor

Date: 12/12/2013

APPENDIX FOR CHAPTER TWO

(vi) Quality assessment scores for each included paper

	Brown et al. (2015)	Burke et al. (2014)	Feros et al. (2013)	Goodwin et al. (2012)	Gregg et al. (2007)	Hawkes et al., (2013. 2014)	Lundgren et al. (2006)	Lundgren et al. (2008)	Nordin & Rorsman(2012)	Rost et al. (2012)	Whittingham et al. (2014)
1. Clarity of sample description	2	0	0	2	2	2	2	2	2	0	0
2. Severity/chronicity of the disorder	-	-	-	-	-	-	-	-	-	-	-
3. Representativeness	1	2	1	2	2	2	2	2	2	1	2
4.Reliability of the diagnosis	-	-	-	-	-	-	-	-	-	-	-
5. Specificity of outcome measures	2	2	2	2	2	2	1	1	1	1	2
6. Reliability and validity of outcomes	1	2	2	2	1	2	2	2	2	2	2
7. Use of blind evaluators	0	0	0	0	0	2	0	0	0	0	0
8. Assessor training	0	0	0	0	0	1	0	0	0	0	0
9. Assignment to treatment	2	0	0	0	2	2	1	1	1	2	2
10. Design	1	0	0	0	1	1	1	2	2	2	2
11. Power analysis	0	0	0	0	2	2	0	0	0	0	2
12. Assessment points	1	1	1	0	0	2	2	2	1	0	1
13. Manualized, replicable, specific treatment programs	2	1	1	1	2	1	2	2	2	1	2
14. Number of therapists	1	0	0	2	1	2	1	0	1	0	1
15. Therapist training/ experience	2	0	2	0	1	1	2	2	2	1	2
16. Checks for treatment adherence	2	0	1	1	0	2	0	0	0	0	1
17. Checks for therapist competence	1	0	1	1	0	2	0	0	0	1	0
18. Control of concomitant treatments	0	0	0	0	0	0	1	0	0	0	0
19. Handling of attrition	2	0	1	0	2	1	1	1	1	2	2
20. Stats analysis and presentation of results	2	2	2	2	2	2	2	2	1	2	2
21. Clinical significance	2	0	1	0	1	1	0	0	0	0	0
22. Equality of therapy hours	0	0	0	0	0	0	2	2	2	2	0
Total quality	**24**	**10**	**15**	**15**	**21**	**30**	**22**	**21**	**20**	**17**	**23**
Total quality AVE	**1.2**	**0.5**	**0.75**	**0.75**	**1.05**	**1.5**	**1.1**	**1.05**	**1**	**0.85**	**1.15**